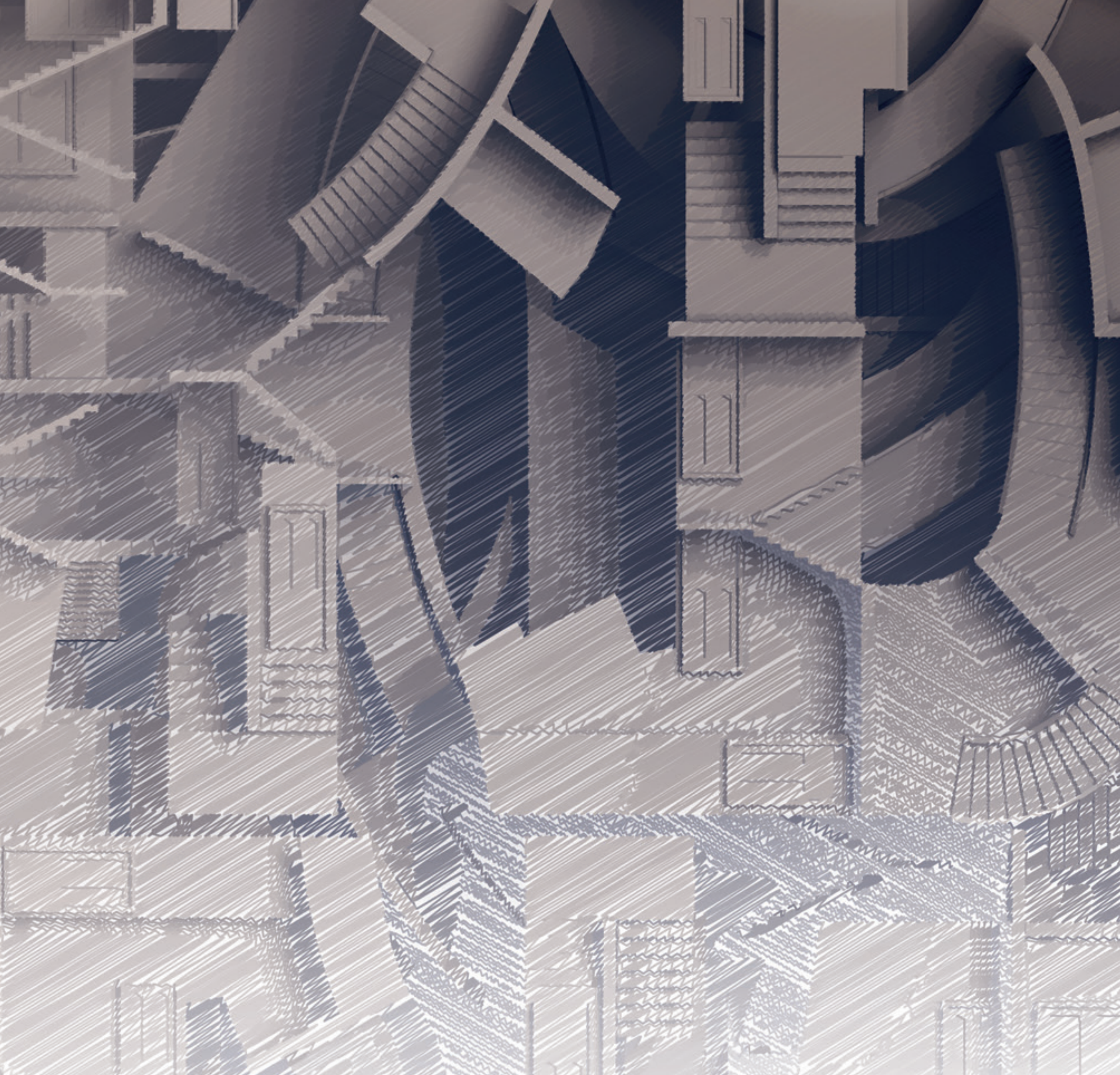

Drawing out the visuomotor abilities of children with mild autism spectrum disorder

DONDERS SERIES

Wim Schlooz

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Cover	Harald Pieper | Promotie In Zicht
Layout	Renate Siebes | Proefschrift.nu
Printed by	Ipskamp Drukkers B.V.
ISBN	978-94-6284-034-8

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Proefschrift

ter verkrijging van de graad van doctor
aan de Radboud Universiteit Nijmegen
op gezag van de rector magnificus,
volgens besluit van het college van decanen
in het openbaar te verdedigen op vrijdag 22 januari 2016
om 12.30 uur precies

door

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geboren op 16 maart 1951
te Venlo

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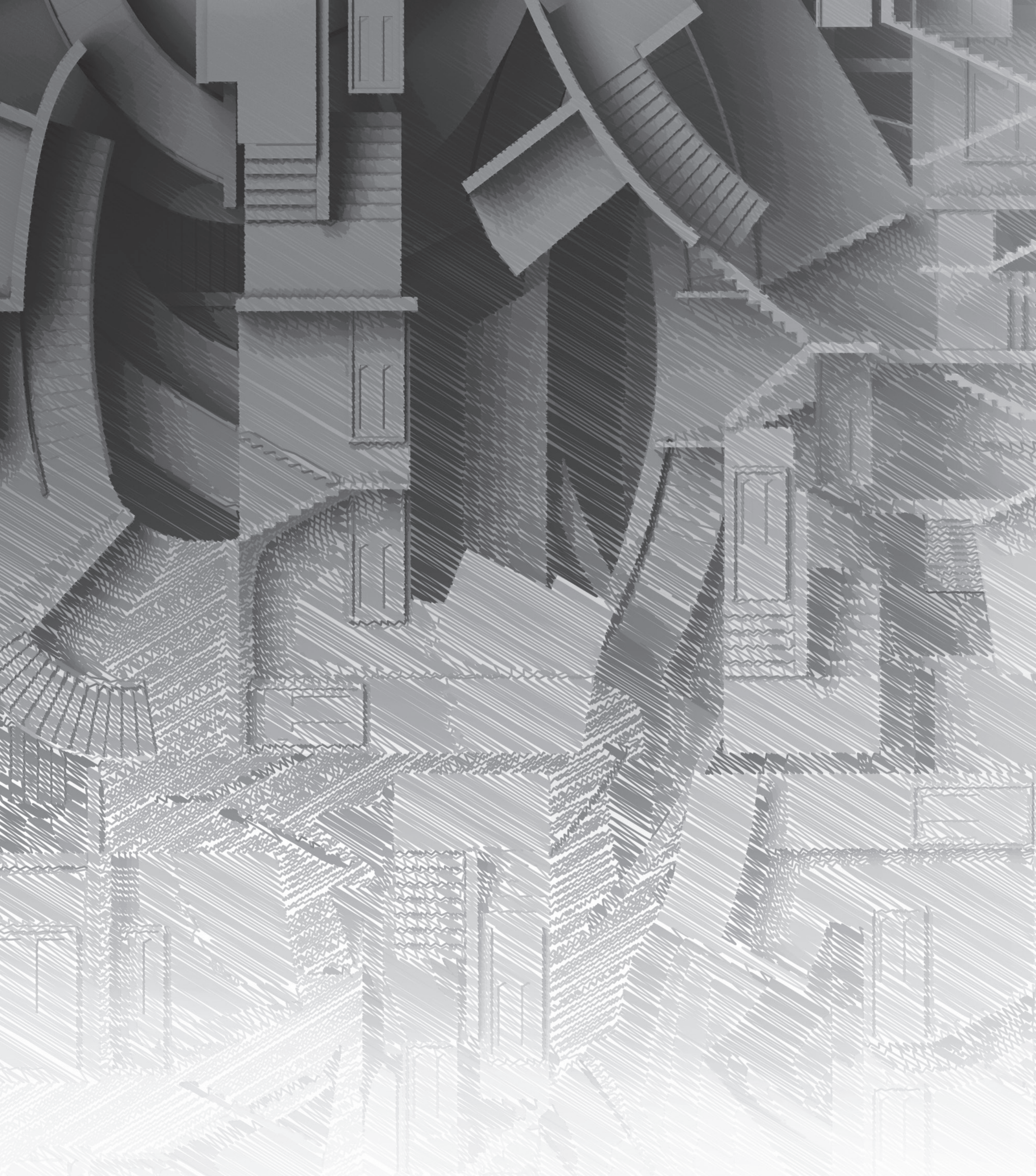

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General introduction and outline of the thesis

Clinicians see numerous children and adolescents with autism spectrum disorder (ASD): they constitute the largest referral group in children's psychiatric outpatient clinics. Both the diagnostic and treatment-management principles have predominantly been derived from and empirically founded on research in children with autism, which syndrome is now referred to as the most severe type within the autism spectrum. Children with severe ASD have great difficulty communicating and engaging in social interactions, while their behaviours and interests tend to be stereotypical (restricted or fixated). Therapists explain these signs to both parents and child to create a greater awareness of the syndrome and the associated (mutual) problems. During this so-called family psycho-education, besides preoccupations and repetitive behaviours, the child's reduced ability to correctly interpret and respond to social cues, i.e. read social situations and generalise experiences, and its unique way of communicating are extensively addressed.

It is, however, debatable whether traditional diagnostic and management practices are also relevant for the much larger group of children and adolescents suffering from a milder type of autism. In this wider ASD group confusion is widespread: recognition of their deficits occurs (much) later and diagnoses are often flawed. In addition to the symptoms described above, in many children struggling with an ASD motor development is atypical or delayed (Fournier, Hass, Nail, Lodha, & Caurugh, 2010), with their motor performance showing distinctive characteristics (Cook, Blakemore, & Press, 2013). The transitions from lying down to sitting up, standing and walking are, for instance, often prolonged and do not transpire spontaneously, while catching a ball tends to remain difficult for quite some time (Asperger, 1944). Children with ASD often take (much) longer to learn to ride a bike or swim and at elementary school tend to lag (seriously) behind in learning to write (Kushki, Chau, & Anagnostou, 2011). To me, as a practising child psychiatrist, it were the problems they experienced drawing a human figure that struck and kept fascinating me.

The process of drawing affords us a wonderful opportunity to investigate the ways visual perception influences human motor control. Prompted by my experiences and ever-growing curiosity, I made the visuomotor functioning of children with mostly milder types of ASD the subject of the research reported in this thesis to gain a greater understanding of the mechanisms underlying the atypical characteristics in their drawings – and to thus help determine whether children with a mild ASD are indeed rightfully treated similarly to their peers with more serious symptoms. To analyse the underlying mechanisms of their deviant drawing performance in more detail, in this thesis we investigate several central aspects of drawing in isolation in subsequent studies: the reproduction of visual details versus global aspects, visual perception and perceptual motor functioning, disembedding capacities and precategorical visual perception.

Even though children with mild ASD constitute the greater group in clinical practice, they have, oddly, received relatively little scientific attention while new systematic insights accordingly have great social relevance. Children and adolescents with mild ASD are also highly researchable: they are very keen to participate in research studies and use relatively little psychotropic or other medication. Psychopharmacological agents in particular may affect reactivity, visual perception and motor performance, all undesirable side-effects when cognitive functioning is the focus of attention.

The main research questions in the studies presented in this dissertation were: Is the visuoperceptual processing of children at the milder end of the autism spectrum different from that of typically developing children and is it indeed characterized by a preference for local over global processing as is known to be the case in children with severe ASD? Are the visuomotor processes distinct from those observed in typically developing children? And what is the role and effect of the supposed atypical visual perception on their motor functioning?

Autism spectrum disorders

Before sketching the background of these queries in more detail, some of the concepts guiding the diagnosis of ASD merit discussion.

The Diagnostic and Statistical Manual of Mental Disorders (DSM) is an important diagnostic and classification benchmark for psychiatrists, comparable with the International Statistical Classification of Diseases and Related Health Problems (ICD). In the previous editions, the DSM-IV (APA, 1994) and the DSM-IV-TR (2000), the autistic disorders falling within the entire severity continuum were categorised and together referred to as 'pervasive developmental disorder' (PDD), which umbrella term included autism and Asperger syndrome, but also Rett's disorder and Heller's syndrome or childhood disintegrative disorder. The remaining disorders were labelled as PDD-NOS (PDD-not otherwise specified). Classifying Rett's and Heller's as a PDD was already considered questionable at the time, as both syndromes concern paediatric pathologies that differ strongly from the other disorders in aetiology and epidemiology as well as course (Lord & Bailey, 2002).

In the DSM-5, the latest edition released in May 2013, the umbrella term 'autism spectrum disorder' (ASD) was adopted, which designation had already been featuring widely in more recent research. The DSM-5 no longer merely classifies and defines the various disorders within the spectrum but now uses a dimensional, i.e. behavioural, developmental and methodical variance approach with differential diagnoses hinging on two key domains: social communication and restricted/repetitive behaviours (Lord & Jones, 2012). At the time of writing this thesis, both the DSM-IV(-TR) and the DSM-5 classification systems prevailed, with all barring the most recent study – presented in Chapter 4 – having been published prior to the latest revision. In this thesis the reader will hence come across designations derived from both classifications, where the children with the severest ASD type are denoted as suffering from autistic disorder (AD) and those diagnosed with milder variants of ASD as having Asperger syndrome or PDD-NOS, with the latter two being labelled as the 'PDD' group.

With the demarcation of autism versus Asperger syndrome and PDD-NOS in itself being cause for much debate, the definition of the outer boundaries of the large cluster of syndromes falling within the current autism continuum has likewise raised many questions. When are symptoms indicative of other clinical syndromes such as AD(H)D, speech-language disorders, or specific disorders like dyslexia, or even typical development? The new ASD definition then has not settled the debate (Lord & Jones, 2012; Kent et al., 2013). In two earlier studies and based on the statistical analyses of the extent to which various symptoms contributed to a sensitive and specific diagnosis in 105 and 285 individuals diagnosed with an ASD, Buitelaar and colleagues (Buitelaar & van der Gaag, 1998; Buitelaar, Van der Gaag, Klin, & Volkmar, 1999) drew up diagnostic rules to help define the boundaries of the autism spectrum. Their rules allowed the distinction between ASD and non-ASD, a highly relevant instrument for both clinicians and researchers. It is these rules that we abided by in the studies presented in this thesis.

The ASD severity continuum

Disorders in the autism spectrum are pervasive and serious. They not only impinge on many functional domains (social contact/communication, speech and language, planning and organisation, and often also motor control), also in every stage of their lives people with an ASD will need to make great efforts to keep up with their age peers, gain and sustain their independence both in living and school/work settings, and maintain fulfilling personal and social relationships, all requiring continued adaptations of and support from their social environment as well as professional guidance.

The cognitive signs and symptoms associated with ASD stand out. A proper understanding of these special features helps the afflicted children and (young) adults, as well as their social environment, in their efforts to hold their ground and make the most of their strengths. Within the spectrum, autism has been the most widely investigated (17,278 publications according to PubMed between 2004 - April 2014). Although far more prevalent, the lesser variants of ASD have received far less systematic attention (1,452 publications in the same period). Yet, it would be wrong to underestimate the problems these children experience. It is children with a milder ASD and their neurocognitive functioning that are the focus of the present work.

Autism or autistic disorder, the severest and core disorder within the spectrum, is a neurodevelopmental syndrome affecting more than one in 600 children. Although clearly delineated in the DSM-IV and having a clear biological basis, and probably constituting the most reliably diagnosed of all paediatric psychiatric disorders (Lord & Rutter, 1995), its diagnosis has always been founded on behavioural observations, i.e. the presence of qualitative impairment in social interactions and communication, and a restricted, repetitive pattern of stereotypical behaviours (DSM-IV, APA, 1994). Dedicated diagnostic instruments were developed for this syndrome, with the Autistic Diagnostic Interview (ADI-R; Lord, Rutter, & Le Couteur, 1994), the Childhood Autistic Rating Scale (CARS; Schopler, Reichler, DeVellis, & Daly, 1980) and the Autistic Diagnostic Observation Scale (ADOS; Dilavore, Lord, & Rutter, 1995) becoming the most widely used, where the ADI-R and ADOS are taken to be the gold standard (Filipek, 1999). However unequivocal the definition of autism may be, much of its pathogenesis remains unclear. From the genetic perspective, for instance, monozygotic twins that both have an ASD have been found to differ as much as 50 IQ points and to lead completely different lives, with one of the twins driving to work by car and its sibling requiring the most basic of care (Lord & Bailey, 2002). More intriguingly, a monozygotic co-twin may even be clinically unaffected. Looking at 20 dizygotic and 28 monozygotic twins of whom at least one had been diagnosed with an autistic disorder, one study found two co-twins in the latter group to function such that no diagnosis was made (Le Couteur et al., 1996).

As stated, children with a disorder at the milder end of the autistic spectrum are far more numerous. In a population study conducted in the UK (Staffordshire) 15,500 children aged between 2.5 and 6.5 years were screened for developmental problems. The survey yielded a prevalence rate of 16.8 per 10,000 children for an autistic disorder and a rate of 45.8 per 10,000 for non-autistic PDD (Chakrabarti & Fombonne, 2001). A more recent review estimated the total number of children with an ASD at an even higher 62 per 10,000, with the male-female ratio ranging from 1.33 to 16.0 for AD and 3.3 to 15.7 for PDD in Europe (Elsabbagh et al., 2012). Supporting epidemiological findings, in clinical practice the children that are referred with a (suspected) milder variant of ASD far outnumber those with autism. In February 2013, the former group constituted 29.4% of the total population being treated at Herlaarhof, the Dutch child psychiatric clinic where I work. In their survey of 770 families raising a child with an ASD, Patricia Howlin and Anna Asgharian (1999) compared families with a child with autistic disorder and those with a child with Asperger syndrome – a milder ASD variant wherein the child does not show a delay in language and cognitive development. They found that the parents with an autistic child started feeling concerned about its development on average 12 months earlier than the 'Asperger parents' (at age 1.52 vs age 2.53), while they also sought help much sooner (at age 2.05 vs 3.49). Their main finding was that, at the time of their survey, the children with autism had been diagnosed at around age 5 and their peers with Asperger's not until age 11. Besides motivating research into the cognitive styles of autistic children, these combined findings reinforce the motive to also do so in children with milder ASDs given that the problems and their remediation proceed along the same lines. Or do they? Are the cognitive patterns in mild ASD indeed similar to those observed in autism?

Neurocognitive accounts of autism

There are three leading neurocognitive theories on autism that have guided research in this field (Happé & Frith, 1996). One of these is the theory of mind account (Baron-Cohen, 1989) that focuses on social communication: people with autism would be less able to mentalise; they are assumed to have little idea about their own and others' mental state (beliefs, desires, intentions), which deficit would lead to impairments in communication, socialisation and imagination, all well-known problems in children with autism.

The two other accounts, the executive function theory and the central coherence theory, emphasise the non-social impairments in children with autistic disorder. Both theories postulate that the social impairment that hampers people with autism is a consequence of underlying non-social cognitive deficits. Executive functions cover a wide array of higher cognitive processes (see Lezak, 1995). Experiments with the Wisconsin Card Sorting Test and the Tower of Hanoi have shown that children with autism have problems with planning and organisation, and that they display a tendency to perseverate (Pennington & Ozonoff, 1996; Russell, 1997). In addition, they are said to use an atypical cognitive style, described as weak central coherence (WCC; Happé & Frith, 1996), which entails that they would make relatively little use of context and pay preferential attention to parts rather than wholes. Earlier, Uta Frith (1989) had assumed that "In the normal cognitive system there is a built-in propensity to form coherence over as wide a range of stimuli as possible, and to generalize over as wide a range of contexts as possible. It is this drive that results in grand systems of thought, and it is this capacity for coherence that is diminished in children with autism." From this hypothesis, the remarkable aptitudes in mathematics, drawing, memory and music 'idiots savants' display (Hermelin & O'Connor, 1990; Heaton & Wallace, 2004) would then be explained. It would also help account for the various other distinctive capacities that have been observed in people with autism: their propensity to be less distracted by visual illusions in experimental settings (Happé, 1996) and to disproportionately remember insignificant details: they are masters in detail, but have little eye for the overall structure. Because of these unique qualities, the term cognitive style is favoured over cognitive deficit (Happé, 1999), with the style being characterised by 'local' rather than 'global' processing, which not only puts people with ASD at a disadvantage; it also is the source of their singular skills. Yet, this cognitive processing strategy does not always prevail: a child that spontaneously shows clear signs of local processing may well be able to shift to a global processing strategy when appropriately instructed (Happé & Frith, 2006).

This atypical cognitive processing style has been investigated in depth. The most illustrative examples are the studies that applied (adaptations of) the Embedded Figures Test (EFT) originally developed by Witkin, Oltman, Raskin, and Karp (1971), which Shah and Frith first used with autistic children in their renowned 1983 study. In their report the authors speak of "an islet of ability," pointing to the superior proficiency of these children to discern details. The adult and children's versions of the EFT are visuoperceptual tests requiring the participant to detect a smaller form that is 'hidden' within a larger figure. Children with ASD recognise this embedded shape better, it is said, because they, unlike typically developing age peers, are less distracted, i.e. delayed, by the meaningful or more abstract context of the larger figure. We have used two adaptations of this test in the studies reported in Chapters 2 and 3.

Prior to this, Hermelin and O'Connor had comprehensively explored neurocognitive functioning in autism, which research they describe in their work entitled *Psychological Experiments with Autistic Children* (1970). Their study contributed greatly to the notion that autism and the associated cognitive and functional impairment did not arise from emotional deprivation (Bettelheim, 1967) but far rather had its origins in neurocognitive deficits. The authors state that at the time of their investigations three theories had already been proposed to explain the phenomena typically observed in autism. One suggested an inability to develop “abstraction,” which would account for the tendency people with autism display “to order the external world and the concreteness of verbal interpretation” (Rimland, 1964). Another deemed the typical restricted or repetitive behaviours to be predominantly acquired, i.e. resulting from “faulty learning” (Lovaas, 1966). The third account interpreted the withdrawal, stereotypical behaviour and diminished pain sensation as a defence against overarousal from too intense stimulation (Hutt, Hutt, Lee, & Ounsted, 1964). Hermelin and O’Connor identified most with Rimland’s hypothesis and scrutinised the sensory and visual perception in children with autism by looking at language, coding, seriation (coherent sorting), recall, and responsiveness. Their main conclusion was that the children are unable “to encode stimuli or to order their environment, meaningfully” (p.129). They posit that the children strongly tend to “impose, stimulus-independent, simple, rigid and repetitive patterns on random as well as structured, meaningful input”, which then results in “an impaired and limited ability for and appreciation and reflection of order, pattern structure and meaning in the environment.” Having co-conducted these experiments and evidently building on their thinking, Uta Frith introduced the weak coherence concept in her later work (1989). Sadly, Hermelin and O’Connor’s paradigms were not all followed up: subsequent research was mainly restricted to visual perception, with far fewer studies being directed at its effects on motor functioning. Since perception is heavily implicated in drawing, we decided to take one of their experiments further in the study described in **Chapter 2**.

Drawing

To learn more about the cognitive style of children with ASD, besides visuoperceptual tests, drawing tasks have been employed (Booth, Charlton, Hughes, & Happé, 2003; Chapter 1), while in the clinical practice of child psychiatry it is not unusual for assessments to also include drawing and writing tests (Trad, 1997). Having observed children with ASD draw, and studying well-over 3000 of their house-tree-person drawings (HTP; Buck, 1948), I could not help but marvel at the problems children with ASD seem to experience when having to draw a person as compared to typically developing children (see Figures 1.1 and 1.2). Likely because of the difficulties they have conceptualising a human figure, they struggle to get the proportions between the details and the total figure right, hesitating where to place and what size to draw them. In the face: eyes, nose, and hair. In the larger body: limbs, hands, fingers, and feet. They also show a remarkable lack of body awareness, finding it very difficult to

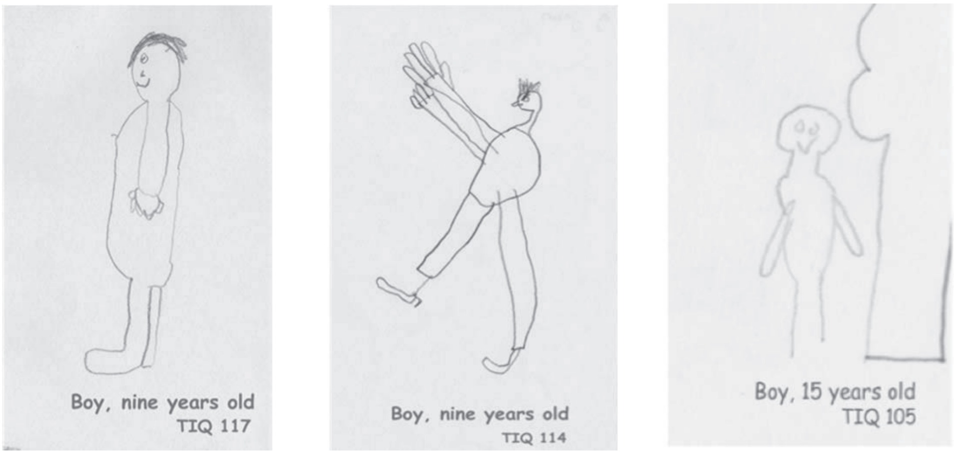

Figure 1.1 Drawings of a human figure, the first two made by children with milder ASD ('PDD-NOS'), the third by an older boy with severe ASD ('AD').

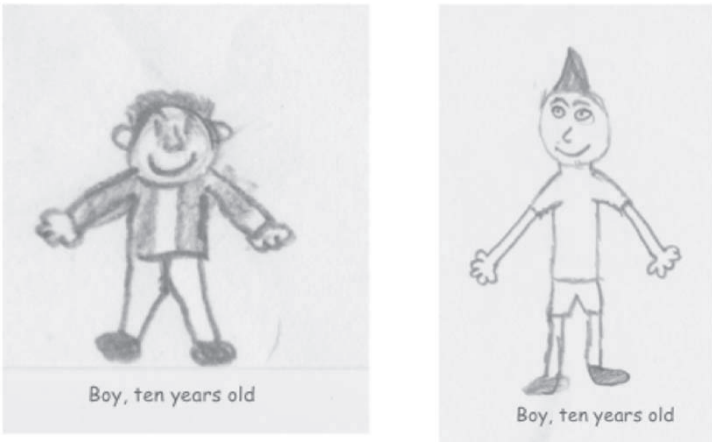

Figure 1.2 Drawings of a person by typically developing boys, aged 10 years.

point to and name body parts, which observations were in agreement with those reported for pre-school children with psychiatric disorders, among which ASDs (Simons, Leitschuh, Raymaekers, & Vandenbussche, 2011).

The question then is whether these atypical representations of human figures are in fact related to the clinical observations of autistic behaviour. To try and answer this question we need to establish what it is that causes children with ASD to have problems drawing human bodies. Van Sommers (1984) mentioned that already when simple figures are being copied, semantic or perceptual factors influence the drawing plans, which might partially explain their ineptness to some degree. Difficulties interpreting emotional states, known as alexithymia and found in some people with ASD (Cook, Brewer, Shah, & Bird, 2014), might also considerably influence their drawing performance. Other studies suggested a reduced ability in so-called prototype formation or categorising (Gastgeb, Rump, Best, Minshew, & Strauss, 2009; Klinger & Dawson, 2001). "Prototype formation is a critical skill for making sense of a world with infinite categories to learn. A prototype is a representation of past information that depicts the average of variations within a category. Forming a prototype decreases memory load allowing individuals to store a single representation of experienced items" (Gastgeb, Rump, Best, Minshew, & Strauss, 2009, p 279). As in children with autism the concept of their own bodies is very poorly represented and developed, their prototyping skills are then also impaired, explaining why they find it difficult to draw a human figure by heart.

Analysing drawing styles

Analysing and comparing the characteristics of human figure drawings of children with ASD and typically developing peers is a very complex task, which is why we restricted our investigations to certain aspects of their drawing performance. Using validated and well-known drawing tasks (see Outline below), we looked at their reproduction capabilities (Rey's CFT and VMI), their assumed preference for global or detail-oriented processing (Rey's CFT, CEFT and EFT), their use of vision in the planning and guidance of their drawing movements, and some aspects of their visual processing strategies (prior knowledge). With the use of these tests we expected to be able to learn more about the atypical cognitive style of children with ASD even if they only suffered from a milder variant of ASD.

To evaluate the drawing performance of our young participants, we used a digital drawing tablet and dedicated software (De Jong, Hulstijn, Kosterman, & Smits-Engelsman, 1996). The technique allows important parameters such as reaction times, drawing time and drawing speed of real-time movements to be recorded, reproduced and analysed offline without distracting the child or interfering with the drawing process. Another advantage of studying drawing is that, apart from the fact that most children love to draw, there is little language in drawing tasks, which is what tends to confuse children with ASD as, in general, they have poor semantic skills and take words or expressions literally. Besides the necessary instructions, we hence tried to avoid verbal communications as much as possible during task performance.

Outline of the thesis

To get a better grip on the deviations children with mild ASD show in visuomotor processes involved in complex skills such as drawing, the study of **Chapter 2** focuses on two aspects. First, to investigate preferences for local or global processing styles, we compared the performance on the Rey Complex Figure Test (CFT) of boys with 'PDD', boys with Gilles de la Tourette syndrome (TS), and typically developing (TD) age peers, with the TS group being included to control for a-specific effects of a neuropsychiatric disorder. To compare the visuoperceptual performance of the three groups, we applied the detailed scoring and analysis procedure developed by Waber and Holmes (1985, 1986, revised 1996), which makes a clear distinction between structural and incidental elements in Rey's CFT, an important distinction when comparing local and global processing styles. In addition, we devised a measure for fragmented processing by quantifying the number of strokes that were used to draw the structural elements. The drawings were digitally recorded and analysed offline without the experimenters interrupting or disturbing the children's drawing activities.

Second, with the Children's EFT we looked for a superior disembedding capacity in the boys with PDD. Were they indeed better able than the boys in the TS and TD groups to find the hidden forms (detail) within the surrounding meaningful figure (global) and was their performance on the Block Design task also superior, as Shah and Frith had earlier (in 1983 and 1993, respectively) found in children with autism?

In **Chapter 3** we once again compare the performance of the same three groups of boys (PDD, TS and TD), this time using two visuomotor tasks in order to learn whether and how the observed differences in visual perception affect their perceptual motor performance differentially. Children and adolescents with ASD-related issues often have problems performing everyday tasks and acquiring daily skills; they, for instance, take longer to learn to catch a ball (Asperger, 1944) or continue to have difficulties writing legibly. We studied the functional domains of visual perception and motor coordination in two tasks: a tracking task designed by Hermelin and O'Connor (1970), which we slightly adapted, and the Beery-Buktenica Visual-Motor Integration test (VMI; Beery, 1967; revised 1997). To selectively study the influence of visual perception on their motor output, the boys completed the tracking task, in which they had to trace a punched-out track, in a blinded and an unblinded condition, with the digitiser allowing us to analyse their performance in detail and offline. The VMI is a standard neuropsychological test assessing the extent to which children can harmonise their visual and motor abilities when required to copy increasingly complex figures, with its subtests gauging the relative contributions of visual perception and motor coordination.

In **Chapter 4** we will be looking more closely at visual perception in mild ASD, thereby contributing to a longstanding discussion. The impairments in motor control children with ASD display in complex tasks like drawing might also be modulated by altered perception. Most findings so far suggest that children with severe ASD perform better on embedded figures tests and might then indeed have "an islet of ability" as Shah and Frith (1983) posited. But is this also the case for children at the milder end of the spectrum? More to the point, do or don't they perform better on the Embedded Figures Test (EFT) than their typically developing counterparts? As explained before, this test played a major role in the conception of the weak central coherence theory in which Shah and Frith (1983) allude to a superior ability, not a deficiency, in children with autism, given that they were better able to find the hidden forms in the surrounding and distracting greater figures than were matched controls. Later results on the test were ambiguous, though. Simmons et al. (2009) discussed all studies published to date in which either the children's, the adult or both versions of the EFT were used and judged the balance of evidence to be in favour of a superior performance of people on the autism spectrum. More recently, White and Saldaña (2011) also reviewed 16 studies using the EFT in people with ASD and likewise found inconsistent results. We concluded (see Schlooz et al., 2006, Chapter 2) that when interpreting the results of studies using an EFT it is important to closely look at the sensitivity of the task: in the earlier studies that employed a children's version there seemed to be a risk of ceiling effects in children with ASD. In the study reported in this chapter (Schlooz & Hulstijn, 2014) we tested this idea by having boys with mild ASD and typically developing boys execute the easier Children's EFT as well as the more complex adult EFT in which a ceiling effect was not probable.

Chapter 5 takes a different perspective. After having mainly reflected on higher-order cognitive skills (using copying and tracing tasks) in the previous chapters, in the study presented in this chapter the focus is on precategorical perceptual processing, visuoperceptual processes that require a lower level of cognition, where task performance is not measured in seconds but in milliseconds thus precluding higher-order processing. We deemed this research angle of great relevance as it is generally assumed that weak coherence mostly applies to higher cognitive processes, the level at which meaning is assigned (Happé & Frith, 2006). In two experiments we explored whether in children with mild ASD ('PDD') visual perception is atypical, i.e. whether they are capable of perceptually integrating structures, also referred to as visual completion, and whether they do so in a contextually dependent manner or by relying more on local processing.

In all our research, we were primarily inspired by the question whether, similar to the patterns observed in children with autism, the cognitive style of children with 'milder variants of autism' is indeed different from that of typically developing children when challenging visuomotor tasks such as drawing are involved, as is often assumed in clinical practice. In this respect, this assumption and our investigations anticipated on the latest views on autism: with the definition of autism spectrum disorder in the DSM-5, all syndromes within the ASD continuum are seen as one cluster, varying only in severity at various dimensions.

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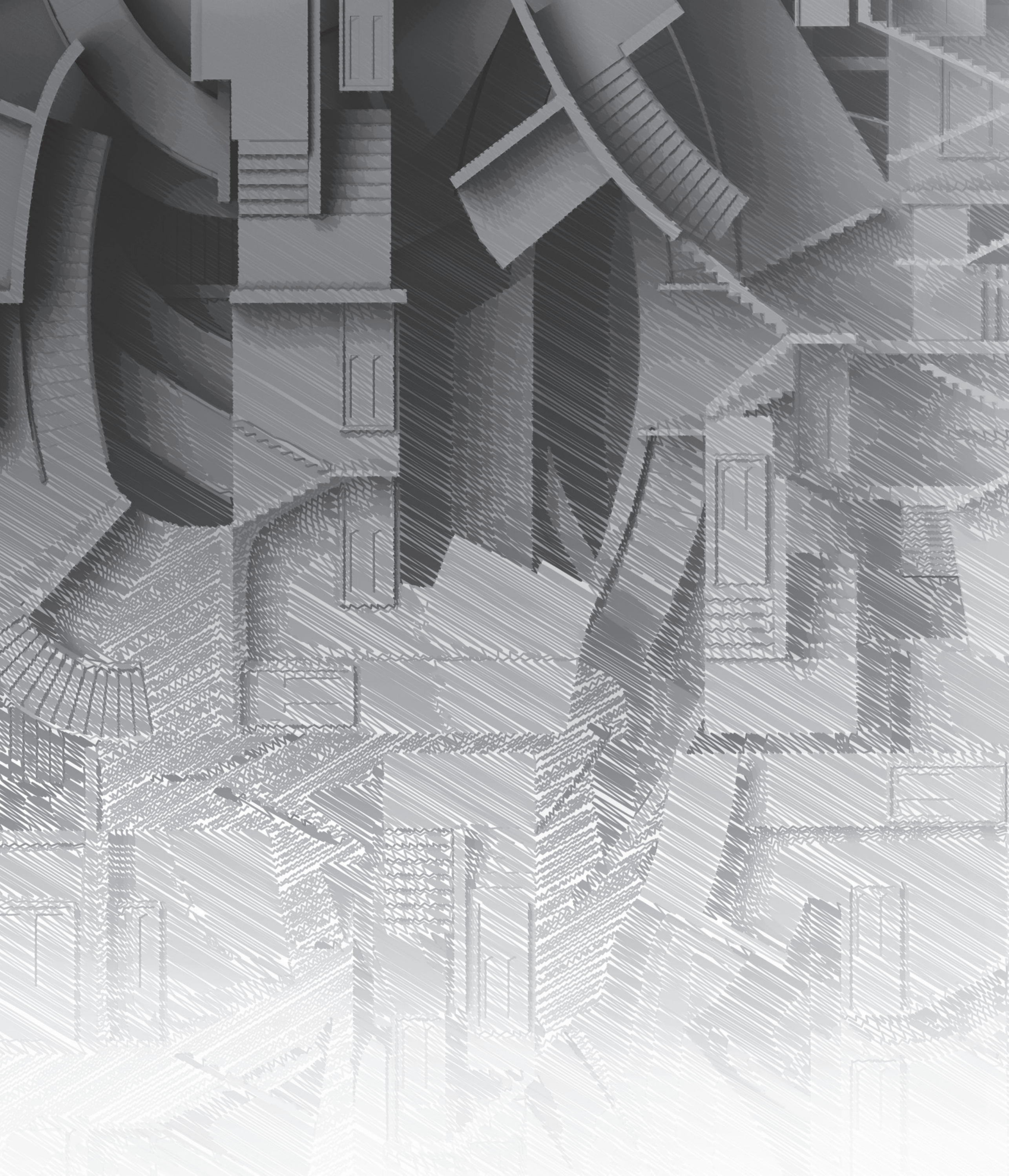

2

Fragmented visuospatial processing in children with pervasive developmental disorder

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Journal of Autism and Developmental Disorders
(2006), 36, 1025-1037

ABSTRACT

Children diagnosed with Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS) and Asperger Syndrome (AS) may be characterised by a similar perceptual focus on details as children with autistic disorder (AD). This was tested by analysing their performance in a visuoperceptual task [the Children's Embedded Figure Test (CEFT)] and a graphic reproduction task [the Rey Complex Figure Task (Rey CFT)]. Control groups were children with Tourette Syndrome (TS) and typically developing children. The TS sample performed similarly to the normal control group in both tasks. The CEFT results did not show the expected preference for local processing in children with PDD-NOS. However, the Rey CFT data revealed that the children with this lesser variant of PDD processed visuospatial information in a fragmented way and were deficient in global processing.

INTRODUCTION

People with autism display an atypical style of perceptual processing. As early as in 1943 Kanner observed that in autism there is "...an inability to experience wholes without full attention to the constituent parts", and this tendency to focus on minute details at the expense of detecting a more global structure, has been recognised by many clinicians after him. Evidence for this atypical perceptual functioning has been found in many studies, the most recent of which being Brian, Tipper, Weaver, and Bryson (2003), Brosnan, Scott, Fox, and Pye (2004; for a review also see Mitchell & Ropar, 2004), Heaton (2003), Mottron, Burack, Iarocci, Belleville, and Enns (2003). However, most of these studies focused on people with autism. Much less is known of the perceptual abilities and deficiencies of children in the larger group of children with Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS) and Asperger Syndrome (AS).

Pervasive Developmental Disorders (DSM IV, APA, 1994) include autistic disorder (AD), AS, Rett Syndrome, Childhood disintegrative disorder and PDD-NOS. Both Rett Syndrome and Childhood disintegrative disorder are clearly different from the other disorders in epidemiology and aetiology. Taken together, the number of children diagnosed with AS or PDD-NOS is much larger than the group with AD: the prevalence rates for the former are estimated at 45.7 per 10,000 preschool children vs. 16.8 for the latter group (Chakrabarti & Fombonne, 2001). As mentioned above, even though far less is known about the perceptual processing in children with lesser variants of PDD, in clinical practice all children within the group diagnosed with PDD are treated as if they were suffering from the same anomalies as are found in AD. It is the aim of the present study to investigate whether this assumption is correct, i.e. whether the population with AS and PDD-NOS, like the group with autism, displays the same preference for a detail-oriented cognitive style and is similarly deficient in global processing.

The term 'PDD' will be used here for the group of children with either AS or PDD-NOS that participated in this study. This use of the term excludes AD, Rett Syndrome and Childhood disintegrative disorder.

People with autism are described to display superior capacities on tasks that favour a detail-oriented cognitive style, particularly in visuospatial tasks. Earlier reports (Shah & Frith, 1983) of a superior performance on the Block Design test and an enhanced aptitude in identifying shapes within a complex picture, as in the Children's Embedded Figure Test (CEFT), led Frith to formulate her influential theory of Weak Central Coherence (Frith, 1989; Happé

& Frith, 1996). According to this theory, people with autism display a weakened tendency to integrate perceptual stimuli into a coherent whole and to generalise over a wide range of contexts (Frith, 1989).

Tests of this theory used tasks like the Block Design test and the CEFT with long-term response times requiring the use of higher-order perceptually categorised information, and involving visuospatial working memory. Later research focused on precategorical perceptual processing, using tasks that are not measured in seconds but in milliseconds, reducing higher-order cognitive processing. These studies focussed on interference between local and global processing in Navon-type tasks (see Navon, 1977; Mottron & Belleville, 1993; Mottron et al., 2003; Ozonoff, Strayer, McMahon, & Filloux, 1994; Plaisted, Swettenham, & Rees 1999; Rinehart, Bradshaw, Moss, Brereton, & Tonge, 2000), disembedding a letter within a task-irrelevant context (Mottron et al., 2003), or visual search tasks (O'Riordan, 2004; O'Riordan, Plaisted, Driver, & Baron-Cohen, 2001; Plaisted, O'Riordan, & Baron-Cohen, 1998).

The findings resulted in alternative accounts for the atypical visuoperceptual processing in children with autism, e.g. an atypical local interference (Rinehart et al., 2000), a local bias (Mottron, Belleville, & Ménard, 1999), an absence of global precedence or a so-called hierarchisation deficit with an enhanced low-level perception (Mottron & Belleville, 1993; Mottron et al., 1999, 2003), an enhanced discrimination ability (O'Riordan & Plaisted, 2001; Plaisted et al., 1999) and an inability to inhibit further processing of irrelevant details (Rinehart et al., 2000). In a recent study Brosnan et al. (2004) point to the lesser utilisation of gestalt grouping principles, leading to a failure to process inter-element relationships. Indications of an absence of global interference (Foxton et al., 2003) and enhanced perceptual functioning (Bonnel et al., 2003) were also found in the auditory domain. However, the exact nature of the deficiency in perceptual processing in autism is not yet fully understood.

The focus of the present study, however, is on the question whether a preference for local processing can also be observed in the larger group of children diagnosed with a lesser variant within PDD. The perceptual style of these children was investigated by employing the CEFT and Rey's Complex Figure Task (the Rey CFT)—both tasks with long-term response times.

The CEFT has become a classic instrument ever since Shah and Frith, in their 1983 study of children with autism, demonstrated superior performance of these children in this task. They were better at detecting the target figures in the complex design. The accuracy scores (number of correct responses) of the children with autism were found to be higher than those of the control groups. There also was a tendency for the clinical group to use a more direct search strategy. Although some of the later studies failed to corroborate the original findings (Brian & Bryson, 1996; Ozonoff, Pennington, & Rogers, 1991), the majority did indeed find a higher accuracy (Ropar & Mitchell, 2001) or shorter response times (Jolliffe, & Baron-Cohen, 1997; Mottron et al., 2003; Ropar & Mitchell, 2001). In addition, in an fMRI study, Ring et al. (1999) showed people with autism to use a different, distinct cognitive strategy in this task.

Rey's Complex Figure Task (Rey CFT) is the second task used in the present study. Here, participants have to copy and reproduce a rather abstract and complex figure. This well-known task is generally used to assess visuospatial planning, as well as constructing and visual memory capacities. The way in which the figure is drawn allows one to infer how the figure is perceived or categorised in structural versus incidental elements and to analyse the sequence in which these elements are being reproduced. Three earlier studies investigated such drawing characteristics with the Rey CFT in children with AD and AS (Jolliffe & Baron-Cohen, 1997; Prior & Hoffmann, 1990; Ropar & Mitchell, 2001). However, the more elaborate scoring and analysis procedure developed by Waber and Holmes (1985, 1986, revised 1996), which makes a clear distinction between Structural and Incidental Elements, was not used in these studies. The Waber and Holmes scoring system allows a qualitative and quantitative analysis of global and local processing in the reproduction of the figure.

In the present study three groups of subjects were compared: children with PDD, typically developing children (TD) and a group with Tourette Syndrome (TS). The latter group was chosen because of a number of similarities between PDD and TS (see also Baron-Cohen & Jolliffe, 1997): children diagnosed with one of these disorders suffer from a developmental disorder from childhood, disrupting their normal schooling and the development of peer relations. According to some authors (Ozonoff et al., 1994; Pennington & Ozonoff, 1996) the two disorders involve frontal abnormalities. They have genetic aetiology and affect males more than females. Therefore, the inclusion of a TS group served to control for a-specific effects of a neuropsychiatric disorder.

To avoid side effects of neuroleptics none of the children selected to participate was taking medication, and to control for age-dependent effects on the CEFT scores the window for chronological and mental age was kept as small as possible: all children were between the ages of 9 and 13, and the groups functioned at an average cognitive level. Because the DSM IV does not provide explicit diagnostic criteria for PDD-NOS, the boundary between the group diagnosed with PDD and non-PDD remains indistinct. In two studies Buitelaar and van der Gaag (1998) and Buitelaar, van der Gaag, Klin, and Volkmar (1999) formulated diagnostic rules for this PDD-NOS category. These criteria and diagnostic rules proved to be more effective in segregating PDD-NOS from non-PDD cases than was possible on the basis of the diagnostic rules prescribed by the DSM IV. These narrower defined diagnostic rules for PDD-NOS, and thus for the whole group diagnosed with PDD, were applied in the present study. It was expected that only children with PDD, and not the children with TS or typically developing children, would show a preference to use a local rather than a global information processing style.

In this study both the CEFT and Rey CFT were administered using special equipment, i.e. a digitising tablet and an electronic pen, which allows precise recordings of the pen movements that participants make when moving towards and pointing to the embedded figure and when drawing Rey's complex figure. This technique, which has hitherto not been employed in the study of visuoperceptual processing in children with autism, ensures a more accurate recording of reaction and drawing times and allows repeated replays of the actual drawing movements, i.e. the sequence in which the elements were put on paper, thus facilitating an accurate and reliable analysis of the children's graphic reproduction behaviour.

METHOD

Participants

Three groups of children, all males, participated in this study: children with PDD, with TS, and typically developing children (TD). The clinical subjects were recruited from regional outpatient clinics for Child Psychiatry associated with the outpatient Clinic of Child Behavioural Neurology and the Academic Centre for Child and Adolescent Psychiatry of the University Medical Centre St Radboud, Nijmegen, the Netherlands. The normal control group was selected from a primary school in a local village. All subjects were functioning on an average cognitive level, without any other disease or illness. None of the participants took medication.

The boys with PDD ($n = 12$) had been diagnosed by experienced clinicians, child psychiatrists and child psychologists both independently and together, using the guidelines of standard criteria such as DSM IV. Three children were diagnosed with AS and nine with PDD-NOS. All children fulfilled the relevant DSMIV diagnostic criteria (APA, 1994). Moreover, we applied the most effective scoring rule for PDD-NOS based on ICD 10/DSM IV criteria

(Buitelaar & van der Gaag, 1998; Buitelaar et al., 1999), i.e. a short set of seven criteria that have all been derived from the original twelve criteria for AD defined in the DSM-IV. The threshold for inclusion in the PDD group was set at three out of seven criteria, of which at least one needed to be in the social interaction domain. The group did not contain any children with autism.

The TS control group included twelve individuals meeting DSM-IV criteria. The selection procedure was identical to that of the PDD group and the diagnoses were made by a child neurologist, a child psychiatrist, a child neuropsychologist and a physical therapist who were all part of a team specialised in the treatment of these children. All children were video-monitored during their visits to the physiotherapist. Children with co-morbidity were excluded.

The mental functioning of all clinical participants was assessed with the Wechsler Intelligence Scale for Children—Revised (WISC-R; Wechsler, 1974). One child with TS only completed a shortened version that, among other elements, did not include the Block Design subtest.

Twelve healthy primary schoolchildren were selected to form the second age-matched control group. As assessed by their teachers and the psychologist, who administered the CEFT and the Rey CFT, they were functioning at a normal cognitive level. Selection was based on an evaluation of the children using a 5-point school-performance scale. Children with a score of either 1 or 5 were excluded from the study.

Table 2.1 presents the chronological age of the three groups and the performance on the Wechsler Intelligence Scale for Children—Revised (WISC-R; Wechsler, 1974) for the two clinical groups.

Table 2.1 Subject characteristics

Group	*N*	Chronological age	Full-scale IQ	Verbal IQ	Performance IQ
PDD	12				
Mean		10;5	105.8	105.3	103.9
Range		9;2–12;10	85–135	81–136	88–125
TS	12				
Mean		11;3	106.7	105.3	107.0
Range		9;5–13;2	83–131	80–122	79–135
Normal	12				
Mean		10;6	Normal range		
Range		9;5–11;5			

Materials and apparatus

By means of a digitising tablet (WACOM 1218 RE) and two specially designed wireless electronic pens, one inking and one non-inking, the drawing movements were digitally recorded. A laptop PC was used to sample and store pen position at a rate of 200 Hz, with a spatial resolution of .2 mm. The recorded drawing movements and drawing characteristics, i.e. response time and stroke sequence, were analysed offline by means of custom-made software (OASIS; de Jong, Hulstijn, Kosterman, & Smits-Engelsman, 1996).

The standard Children's Embedded Figure Test (CEFT; Witkin, Dyk, Faterson, Goodenough, & Karp, 1971) consists of 25 colourful, complex figures in eleven of which the simple outline of a 'Tent' is embedded and in the other fourteen the more complicated form of a 'House' (Figure 2.1). During the instruction procedure and the practice trials the children were allowed to use two cut-out cardboard models of the 'Tent' and the 'House' representing the hidden forms, but they were kept out of view during task performance. The stimulus pictures, each measuring 18 × 24 cm, were coated with a plastic foil to prevent traces. The sheets were presented on the digitiser that was placed at approximately 30 cm from the children's eyes. In this task the children used a wireless non-inking electronic pen to trace the embedded figures.

In Rey's Complex Figure Task (Rey CFT) a drawing of an unfamiliar, abstract nature (the complex figure, see Figure 2.2) measuring 15 × 20 cm had to be copied or drawn from memory (with and without delay) on a blank sheet of paper that was also placed on the digitising tablet. Three sheets of paper were used in each test—one for each condition. Here, the children also produced their drawings with a wireless electronic but this time inking pen.

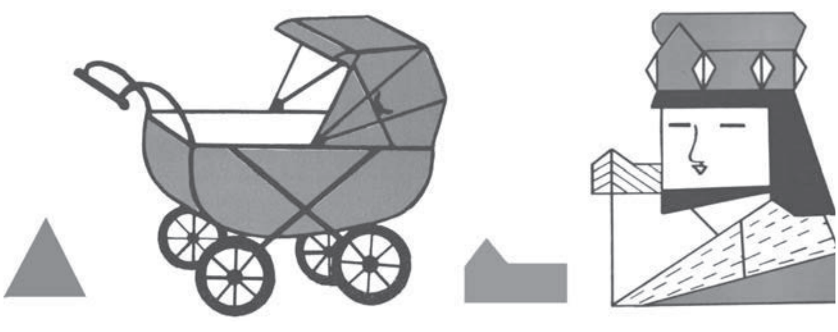


Figure 2.1 The Children's Embedded Figure Test; examples of a 'Tent' and a 'House' condition. The embedded figures are displayed at the left side of the pictures.

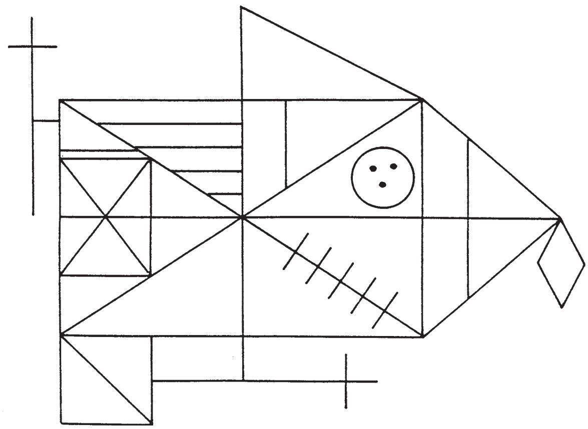


Figure 2.2 The Rey Complex Figure.

Procedure

The two clinical groups were tested in the hospital. The non-clinical controls were assessed at their own school. The tests were supervised by three experimenters working in pairs, one instructing and assisting the child and the second operating the equipment and coordinating the recordings. Due to the digital recording technique interference by the experimenters was reduced to a minimum. Each child was tested individually, and the sequence of the tasks was the same for each child.

The CEFT was presented in accordance with the original procedure (Witkin et al., 1971): when they were instructed and during the practice trials the children were shown the cut-out target shapes to allow them to familiarise themselves with the embedded figures. During task performance, however, the models were put away. The children were instructed to try and find the embedded shape as quickly as possible, and to subsequently trace the embedded form in the complex picture as accurately as possible using the electronic pen. The stimuli containing the embedded 'Tent' shapes were presented first, followed by the stimuli containing the 'House'. The dependent variables were the number of correct responses (accuracy) within the time allowed and the mean response time, analysed separately for the 'Tent' and the 'House' tasks.

In the second task the children were asked to reproduce a complex figure, the Rey CFT (Figure 2.2), on a blank sheet of paper, using the inking pen, in three conditions, i.e. a Copy, an Immediate Recall and a Delayed Recall condition. After having copied the figure in the Copy condition, the children were requested to make a second drawing, but this time the stimulus figure was removed and they had to draw the picture from memory (Immediate Recall condition). Following the Immediate Recall condition the children carried out a non-visuospatial task, i.e. a word categorisation task (the data of which will not be reported here) for 15 min, after which they were once again asked to draw the Rey design from memory (Delayed Recall condition).

Data analysis

CEFT response times were measured from the time a stimulus picture was presented until the pen started to trace the detected 'Tent' or 'House'. If initially a wrong shape was detected that was subsequently corrected, then the start of the corrected tracing was taken as the response time.

Two extra response times were calculated to allow a comparison with findings from previous studies (Jolliffe & Baron-Cohen, 1997; Ropar & Mitchell, 2001). The first included the errors, i.e. the response times for the wrongly traced figures, and the second also included the trials where the subject had failed to find the embedded figure. In accordance with Jolliffe and Baron-Cohen (1997), and with Ropar and Mitchell (2001), the response time in these missing cases was set at 180 s. These three different response time measures were calculated for all items and, in order to get a more reliable mean response time, on a subset of relatively easy items, i.e. the five 'Tent' items and the seven 'House' items with a percentage of correct answers of 80% or higher. Thus, six alternative response times were calculated.

The results of the CEFT were analysed by means of a GLM Repeated Measures procedure with Condition as within-subject factor (Tent versus House) and Group as between-subject factor (PDD versus TS versus Controls). Two separate GLMs were conducted to evaluate the percentage of correct responses and the mean response times. In all analyses *P*-values lower than .05 were considered to be significant.

The Rey CFT drawings were analysed separately and scored offline according to the scoring criteria developed by Waber and Holmes (1985, 1986, revised 1996). These criteria are based on four scoring principles, viz. Organisation, Style, Accuracy and Errors.

The *Organisation* score is a weighted score of the number of structurally important elements that are present in a subject's reproductions, i.e. the number of alignments and intersections of the figure, like the four corners, and other important parts of the base rectangle, e.g. the smaller left center box, and the central crossing point (see Figure 2.3).

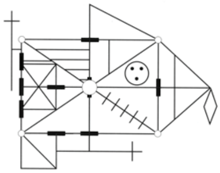
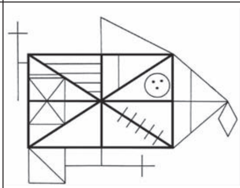
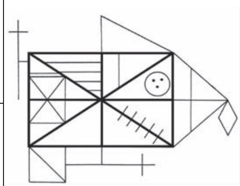
Term	Scoring principle for raw scores	Illustration
Organization	Counting alignments (bold) and intersections	 (Immediate Recall Condition)
Style	Continuity of drawing (bold parts only)	 (Immediate Recall Condition)
Accuracy Structural Elements	Number of Structural (bold) Elements	
Accuracy Incidental Elements	Number of Incidental (thin) Elements	

Figure 2.3 Scoring principles Rey CFT.

The Style scoring principle is defined by the lines that constitute the Base Rectangle, and the intersecting lines within, bisecting the rectangle in horizontal, vertical or diagonal parts (Figure 2.3). Depending on the way these lines are drawn, continuously or fragmented, each reproduction is classified as being drawn in a Configurational, Intermediate or Part-Oriented style.

The *Accuracy* score represents the total number of elements drawn. The score is divided into the subscores Structural and Incidental Elements. The Structural Elements consist of the constituting lines of the Base Rectangle and the intersecting lines within. All the other elements inside and outside the rectangle are considered to be structurally less important and are called the Incidental Elements (Figure 2.3).

Error scores are obtained by summing the number of Rotations, Perseverations, Misplacements and Conflations of each reproduction. Because the number of errors is highly influenced by the number of elements a child reproduces, error rate is expressed as a percentage of the total number of elements reproduced.

In addition to the Waber and Holmes scoring criteria, a *Fragmentation* score was employed in this study to further quantify the children's drawing styles. This score reflects the number of strokes that were used to construct the Structural Elements (i.e. base rectangle and intersecting lines) of the complex figure. A stroke was defined as the interval between two decelerations. The total number of strokes the child used was then divided by the number of structural elements that were actually drawn (i.e. the 'Structural Element Score').

The Rey CFT results were analysed using three different techniques. For the Organisation Score and Fragmentation Score a GLM Repeated Measures analysis was applied with Condition as within-subject factor (Copy versus Immediate recall versus Delayed recall) and Group as between-subject factor. Since Style scores could have three categorical values only, they were analysed with a χ^2 test. Because Accuracy and Error scores could have somewhat higher numbers but had no normal distribution, an analysis with a non-parametrical test (Mann–Whitney *U*-test) seemed more appropriate.

RESULTS

Children's Embedded Figure Test

The percentages of correct responses and the response times are displayed in Table 2.2. The percentages of correct responses were relatively high in all three groups, and there were no differences between the groups ($F(2,33) = .441$, $P = .647$). However, there was a significant difference in mean percentage correct between the Tent and House conditions ($F(1,33) = 7.602$; $P = .009$).The interaction between Group and Condition was not significant ($F(2,33) = .180$, $P = .836$). The mean total number of correct responses of the PDD group (19.83) was not larger than that of the combined control groups (20.90; $F(1,34) = .624$, $P = .435$).

Response times were calculated in six different ways (see data analysis section). Table 2.2 displays the mean values of the purest alternative in which the response time measure was less influenced by errors, i.e. the correct or corrected response times of those items of which the percentage of correct answers was 80% or higher. There was a significant difference in

Table 2.2 Children's Embedded Figure Test. Mean (SD) percentages correct responses and mean (SD) response times (RT) in secs, in the CEFT per group and condition

	% Correct Tent	SD	RT Tent	SD	% Correct House	SD	RT House	SD
PDD	86.4	17.1	4.07	1.21	73.8	18.1	6.64	1.69
TS	92.4	8.5	5.61	2.45	82.1	16.3	8.46	3.48
Controls	85.2	20.1	4.60	2.04	76.8	17.0	6.76	2.79

this measure between the 'House' and the 'Tent' condition ($F(1,33) = 41.92$, $P < .001$) but the groups revealed no significant differences ($F(2,33) = 2.127$, $P = .135$) and the Group by Condition interaction was not significant either. The five additional response time measures produced quite similar results. The detection of the 'Tent' was always significantly faster than the detection of the 'House', but none of the group differences was significant. Even when the two control groups were combined, the mean correct or corrected RT on selected items of the PDD group (5.357 s) was not significantly shorter than the corresponding mean RT of the control children (6.357 s; $F(1,34) = 1.749$, $P = .195$).

Rey Complex Figure Task

Table 2.3 presents the group means of the Organisation and Error scores on the Rey CFT for the three groups separately. However, because all the scores of the TS group and the Control group were very much alike and because statistical tests did not reveal any significant differences, in the statistical analyses these two groups were pooled into one control group, which was then compared to the PDD group. Before presenting the results on the separate scores of the Rey, the results of a multivariate analysis on five of the Rey variables (i.e. Organisation, Accuracy—number of Structural and Incidental Elements-, Errors, and Fragmentation) are presented. This analysis, in which the scores were averaged over the three conditions (Copy, Immediate Recall and Delayed Recall) resulted in a significant difference between the PDD group and the two controls groups ($F(5,30) = 4.838$, $P = .002$).

Organisation

The Organisation scores for the three groups are displayed in Table 2.3. It is clear that the PDD scores are lower than the scores of the control groups, especially in the recall conditions. The TS and Control scores are almost equal. The results were analysed using a GLM Repeated

Table 2.3 The Rey Complex Figure Task. Mean Rey CFT Organisation scores (SD) and % Errors per Element, per group in the three different conditions

		Copy	SD	Immediate Recall	SD	Delayed Recall	SD
PDD	Organisation scores	8.0	(4.1)	4.5	(3.7)	4.8	(2.7)
	% Errors per element	3.1	(4.2)	5.1	(4.6)	10.6	(7.0)
TS	Organisation scores	10.3	(1.4)	9.5	(3.6)	10.2	(3.5)
	% Errors per element	1.1	(1.0)	4.7	(4.8)	4.0	(3.5)
Controls	Organisation scores	11.2	(2.9)	8.8	(4.2)	9.1	(4.1)
	% Errors per element	.5	(1.0)	3.1	(3.5)	3.0	(3.7)

Measures, with Group (PDD versus Combined Control group) as between-subjects and Condition (Copy versus Immediate Recall versus Delayed Recall) as within-subject variable. The analysis yielded a significant main effect for the Group variable ($F(1,34) = 18.131$, $P < .001$). There was also a significant effect for Condition ($F(2,33) = 7.866$, $P = .002$). There was no significant interaction effect ($F(2,33) = 1.316$, $P = .282$).

Style

Table 2.4 shows the distribution of the three Style categories across the three groups for all three conditions, leading to a total of 36 drawings for each group. For the PDD group, 'part-oriented' is the predominant style classification, while in the other two groups the most frequently used style of drawing is 'configurational'. When two or three of the reproductions were scored as 'part-oriented', we categorised the drawing style of these children as 'part-oriented'. This criterion applied to 58% of the PDD group and to 29% of the two control groups ($\chi^2 = 2.864$, df = 1, $P = .046$, one-sided). For the Copy condition the corresponding data were 50 vs. 17% ($\chi^2 = 4.431$, df = 1, $P = .018$, one-sided) suggesting that this style difference was already evident in the first condition.

Table 2.4 The Rey Complex Figure Task. Total amount of drawings for the Rey CFT for each group in the three different Styles

	Configurational	Intermediate	Part-oriented
PDD	8	10	18
TS	15	9	12
Control	16	10	10

2

Accuracy

The Accuracy scores for Structural and Incidental Elements are shown in Figure 2.4. The scores are displayed as percentages of the maximum score obtainable. The PDD group differed significantly from the Combined Control group in each of the three conditions on accuracy in reproducing structural elements (Copy: $Z = -2.521$, $P = .006$; Immediate Recall: $Z = -2.948$, $P = .002$; Delayed Recall: $Z = -2.575$, $P = .005$). For the Incidental Elements no differences were found ($P > .10$).

Errors

Table 2.3 also shows the number of errors (in percentages) produced by each group for each condition. The analyses revealed differences between the PDD group and the Combined Control group in the Copy condition ($Z = -1.643$, $P = .050$) and particularly in the Delayed Recall condition ($Z = -3.314$, $P < .001$), but not in the Immediate Recall condition ($Z = -.780$, $P = .218$).

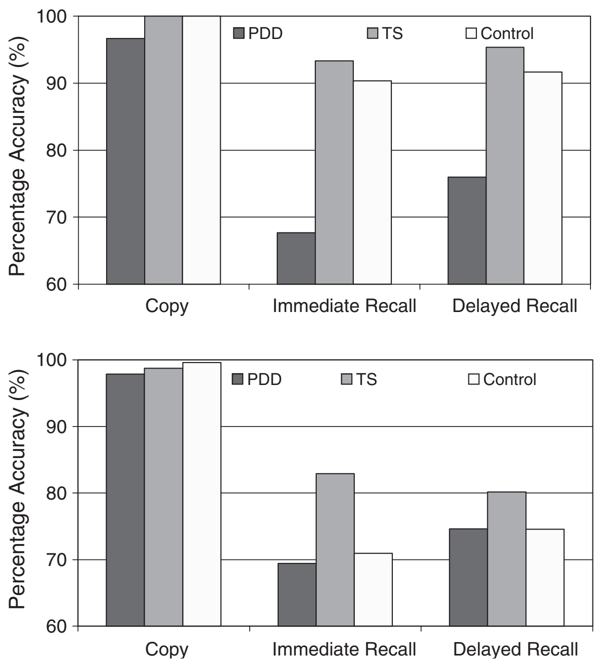


Figure 2.4 Mean Rey CFT Accuracy as the number of reproduced Structural (upper figure) or Incidental Elements expressed as percentage of the total amount of Structural or Incidental Elements per group and condition.

Fragmentation

Fragmentation scores are displayed in Figure 2.5 as a quotient of the number of strokes used to draw the structural elements and the total number of structural elements (= 'Structural Element Score') actually drawn. Analyses with Group (PDD versus Combined Control group) as between-subjects and Condition (Copy versus Immediate Recall versus Delayed Recall) as within-subject variable yielded only a significant main effect for Group ($F(1,33) = 15.304$; $P < .001$). The effects of Condition ($P = .278$) and Group by Condition ($P = .566$) were not significant.

Block Design

From the Full Scale WISC-R, the scores on the Block Design (BD) subtest were derived of the 12 children with PDD and 11 children with TS, and subsequently correlated with the CEFT and the Rey CFT scores. The mean BD score of the PDD group (11.25) was not significantly higher than that of the TS group (10.64; $F(1,21) = .24$, $P = .629$). Therefore the correlation with CEFT and Rey CFT scores was calculated on all PDD and TS children. No significant correlations were found between BD and percentage correct and mean RT of the CEFT ($r = .09$ and $r = -.32$, respectively). However, with the Rey CFT scores a few interesting correlations were found. The Organisation scores did not correlate significantly with BD, but the Style scores did. Children with a Part-oriented Style ($n = 9$) had higher BD scores (12.67) than children ($n = 14$) with an Intermediate or Configurational Style (9.86; $F(1,21) = 6.148$, $P = .022$). In addition, many children, particularly in the TS group, were relatively more accurate on Structural than on Incidental Elements. When the difference

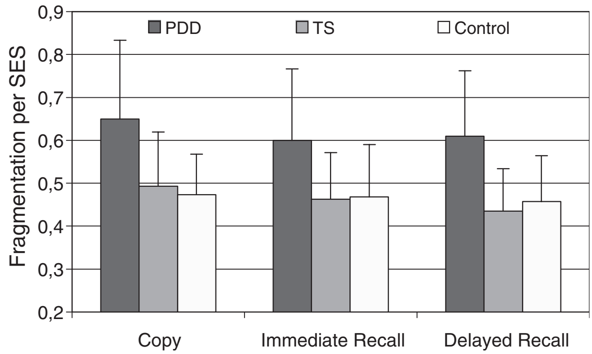


Figure 2.5 Rey CFT Fragmentation scores (+SD) per Structural Elements score as a proportion of the maximal score (=1.0), per group and condition.

between Structural and Incidental scores was correlated with the BD score, significant correlations were found in Immediate Recall ($r = -.402$, $P = .029$), and in Delayed Recall ($r = -.408$, $P = .027$). The Rey Error scores did not correlate significantly with the BD scores but the Fragmentation score in the Copy condition, not in the Recall conditions, and the BD scores did reveal a significant correlation ($r = .46$, $P = .013$).

GENERAL DISCUSSION

In the present study an assumed tendency for detailed visuospatial information processing of children with PDD was assessed in two quite different tasks: a task requiring perceptual recognition of elementary spatial figures (CEFT) and a task in which graphic figures have to be reproduced either in a direct copy condition or from memory (Rey CFT). Contrary to our expectations the CEFT findings failed to show a superior performance of the PDD group relative to the two control groups, both for correct responses and for response time. On the other hand, the performance on the Rey CFT did reveal significant differences between the PDD and control groups. The children with PDD had poorer scores for Organisation, employed a more Part-oriented drawing style, reproduced fewer Structural Elements, made more errors, and had considerably higher scores for Fragmentation.

Children's Embedded Figure Test

On the CEFT no group differences were found, neither in the percentage of correct responses nor in response times. In general, the performance of all three groups was of high quality. Although the detection rate of the more difficult figure ('House') was significantly inferior to that of the simpler triangle ('Tent'), yet in both conditions the percentages of correct responses were quite high (ranging from 73.8 to 92.4%).

Normative mean scores for the CEFT are available for two age levels, i.e. 16.6 (SD = 5.4) for age 9–10, and 18.9 (SD = 5.5) for age 11–12 (Witkin et al., 1971). Only four children in our study scored below the expected mean. Three children with PDD, 6 with TS and 4 TD children scored in the upper range of 23 to the maximum of 25. Therefore a lack of group differences cannot be attributed to a pure ceiling effect, but the fact that all groups scored considerably higher than the norms seems to indicate that this test was not sensitive enough to distinguish subtle group differences. In addition, although the response times in the 'House' condition were significantly prolonged relative to those in the 'Tent' condition, overall the group means were rather low (i.e. 4–5 s) compared to the response times found in other studies, again suggesting that this task was relatively easy for the children participating in the present study. In sum, the task may indeed not have been difficult enough to allow subtle group differences to become manifest.

It should be noted that not all earlier studies found evidence of a superior detection of the embedded figure in people with autism. Although Jolliffe and Baron-Cohen (1997), Mottron et al. (2003), Ropar and Mitchell (2001) and Shah and Frith (1983) reported positive results, Brian and Bryson (1996), and Ozonoff et al. (1991) could not corroborate these results. However, the latter two research groups had, apart from participants with AD (12), also included subjects with AS (1) and PDD (5).

Several connotations regarding this discrepancy in findings need to be made. Firstly, the diagnostic category of the children in our study has to be taken into consideration: it comprised children with PDD-NOS (n = 9) in addition to children with AS (n = 3). Possibly, children with PDD-NOS might display the perceptual style of children with AD to a lesser extent.

Moreover, there are differences in type of tasks and task procedures between the various studies. In some studies the Children's version of the EFT was used, whereas in others the more adult version of the EFT was applied or even specially designed alternatives. In addition, the embedding surroundings varied between meaningful, abstract and fragmented. Also, the method of presentation and availability of the cut-out models of the embedded shapes before and/or during task performance, as well as the size of the surrounding figures, varied. These dissimilarities make it difficult to find a single cause for the contrasting findings in the various studies, including the study reported here.

Of the three studies that used the Children's version of the EFT only the study by Shah and Frith (1983) revealed superior figure detection in children with autism. However, the children in this study functioned at a retarded mental age. In the two studies that failed to find similar results, the participating children were of normal intelligence (Ozonoff et al., 1991) or of a higher age (Brian & Bryson, 1996). This suggests that the superior performance of children with PDD only manifests itself if the task difficulty is sufficiently high to challenge the controls. This would explain why Ropar and Mitchell (2001) with the use of the adult version of the EFT (form A) found group differences and why Jolliffe and Baron-Cohen (1997) with the EFT found faster disembedding performance in AD and AS subjects aged between 19 and 49 years.

This reasoning strongly suggests that the findings of the present study might result from a ceiling effect due to the fact that the CEFT was too easy for the participants. Therefore, they do not allow firm conclusions to be drawn.

Rey Complex Figure Task

The second task used in this study, the Rey CFT, provided a completely different picture. The Organisation scores on this test were lower in the group with PDD than in the two control groups, especially in the two recall conditions.

The Style scores in the various conditions suggested that the children with PDD used a more part-oriented and a less configurational drawing style than the control groups. It is important to note that these group differences in organisation and style were not only evident in the recall conditions but were also manifest in the Copy condition.

In the Copy condition this difference in style was not accompanied by a lack of accuracy, whereas in the recall conditions the Accuracy scores did reveal interesting effects. The controls were able to recall the structural elements quite accurately (scores over 90%), while the PDD group had much lower scores. The recall of incidental elements was lower (i.e. about 75%) in all groups, and did not reveal any group difference. Probably the PDD group did not make a distinction between the Structural and the Incidental Elements. They might perceive the structural elements as merely incidental. If no structure is discerned then the number of elements to be recalled is higher, which will influence the accuracy in the recall conditions.

In addition, the number of correctly reproduced elements, as reflected by the Error score, was lower in the PDD group, particularly in the Delayed Recall condition. This indicates that the encoding of the reproduced elements was of significantly poorer quality for the group with PDD.

The Fragmentation scores demonstrate what remained hidden in the Style scores: the PDD group used more line segments to draw the elements. And they did so from the very beginning, i.e. in the Copy condition, without further deterioration or amelioration in the two recall conditions. This fragmented style of drawing also explains why the Organisation scores of the group with PDD were lower than those of the control groups. Good examples of the fragmented style of drawing in PDD are presented in Figure 2.6. It depicts the interpretations of the Rey CFT by two boys with PDD-NOS (age yr 10;3, WISC-R: TIQ 98; VIQ 99, PIQ 98 and age yr 10;5: TIQ 116; VIQ 116 and PIQ 111) in the Immediate Recall condition and

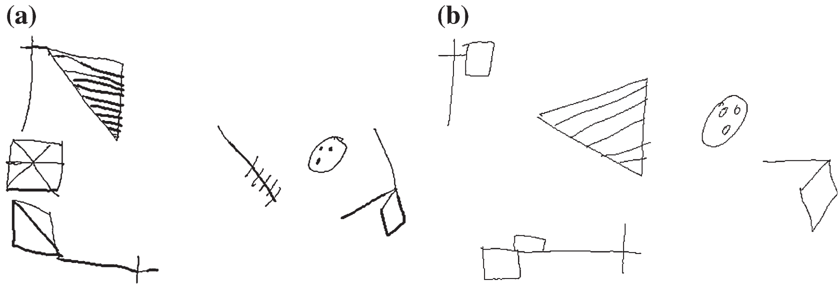


Figure 2.6 Two examples of how the Rey Figure was drawn by children with PDD in the Immediate Recall condition.

shows how much of the quantitative and qualitative information of the original figure was lost. The extent of the contribution to the Rey CFT results of low-level perceptual processes like visual search and interferences between local and global features remains concealed by higher-order processes such as remembering, planning and reproduction. The Accuracy scores seem to point to differences in higher-level perceptual processes between the children with PDD and the control groups.

In general, the results on the Rey CFT are in agreement with the earlier studies in which the Rey was administered in individuals with autism (Jolliffe & Baron-Cohen, 1997; Minshew & Goldstein, 2001; Prior & Hoffman, 1990). Also, Prior and Hoffman (1990) reported that children with autism display a tendency to recall details and not the outline of the figure. Jolliffe and Baron-Cohen (1997), using a modified and simplified figure, noted a trend in both adults with autism and those with AS to use a more local strategy, i.e. to draw the details first and completing the outline later. In addition they reported a more fragmented way of drawing, i.e. the subjects used more lines to complete the figure—which is in line with our current Fragmentation scores. The conclusion however, that a participant starts drawing a detail first before drawing the outline, is fraught because a particular stroke can be part of a detail as well as part of a global structure of the drawing. In this study the preference for a local- versus a global-oriented reproduction strategy was uncovered by differentiating the Rey figure into Incidental and Structural Elements and by counting the number of elements that were reproduced for each class. Minshew and Goldstein (2001) found no effects in the copy condition, but only in the immediate and delayed recall conditions in which more elements of the Rey figure were lost in the group with autism. There is only one study on the Rey CFT in autism in which negative results are reported (Ropar & Mitchell, 2001). The authors failed to detect a significant difference in the use of a local versus global drawing strategy. However, their definition of a 'local' drawing style may have been too broad, because, contrary to the present study, they did not use the more refined scoring procedure proposed by Holmes, Bernstein, and Waber (1996) and Waber and Holmes (1985, 1986) but had two raters provide a judgement on the use of 'fragmented' drawing strokes and on 'incorrect spatial arrangement of the parts of the design'. The combination of the detailed Waber–Holmes scoring procedure and the digital recordings of the drawings, which method avoids the interference of changing crayons and allows elaborate offline analyses, might have contributed to the positive results of the study presented here.

Similar results as those we report here were also found in studies on copying and drawing performance using figures other than Rey's CFT. Mottron et al. (1999) asked autistic individuals to reproduce possible as well as impossible figures and noted that autistic individuals "did not favour global features as much as controls did at the beginning of their production" (p. 749) and "produced more local features at the start of copying". In the discussion Mottron et al. (1999) could not decide between two alternative interpretations of their results: a deficit in perceptual or in executive processes. Booth, Charlton, Hughes, and Happé (2003) tried to disentangle a perceptual from an executive dysfunction (see Russell, 1997) by asking the children to copy a figure and subsequently to draw the same figure but now with a number of specific details added. The latter condition allowed for an extra score measuring advance planning. Poor planning of the group with autism spectrum disorder was found to be unrelated to a detailed style of drawing, suggesting that the two interpretations in terms of a perceptual and an executive deficit can be true at the same time.

Block Design

It was surprising that the PDD group did not differ from the group with TS with respect to the BD scores. Although Mayes and Calhoun (2003) recently also failed to find high scores on the BD in a group of high-functioning children with AD, Goldstein, Beers, Siegel, and Minshew (2001) did report high BD scores in a group of high-functioning adults with autism. In the present study no correlations were found between BD and the CEFT scores, but a significant correlation between the Rey CFT and BD was established for those scores that signal a detail-oriented cognitive style, i.e. a Part-oriented Style, the Fragmentation score and the difference between Structural and Incidental Elements. However, it must be noted that the groups in the present study were rather small.

Tourette Syndrome

Our group of children diagnosed with TS displayed no dysfunctions: they performed at the same the level as the non-clinical controls in each of the tasks and in every condition. Deviant scores were only found in the group of children with PDD and can thus be said not to be a-specific features of neuropsychiatric disorders. The selection procedure for our TS group was such that it excluded co-morbid PDD and Obsessive–Compulsive Disorder. In other studies that assesssed execution tasks and Rey's CFT with the Waber–Holmes organisational method (Harris et al., 1995; Schuerholz, Baumgardner, Singer, Reiss, & Denckla, 1996; Schultz et al., 1998) the TS samples without comorbidity also performed comparable to the normal control groups. Ozonoff et al. (1994) compared children with TS to children with AD and non-clinical controls. For their executive task they used a Go/No-Go paradigm. And again the scores of the TS group were similar to those of the 'normal' controls. However, when accompanied by co-morbid disorders, executive dysfunctions are prominent in TS (Ozonoff, 1997; Ozonoff, Strayer, McMahon & Filloux, 1998).

The present study focussed on children with lesser variants of PDD (i.e. PDD-NOS and AS) and not on children with autism (AD) that are generally the focus of investigation. It must be stressed that our PDD group was selected based on the narrow defined diagnostic rules for PDD-NOS as defined by Buitelaar and van der Gaag (1998) and Buitelaar et al. (1999). Moreover, we used a small window of chronological and mental age for our participants to avoid developmental interference. These facts have to be taken into account when interpreting the current results. Although interesting, the topic of how and when the presentation of a cognitive style develops is not addressed in this study.

There is no doubt about the existence of a specific kind of dysfunction or peculiar cognitive style in this particular PDD group, a dysfunction similar to that described for children diagnosed with a typical AD. Filipek et al. (1999), in their review of neurocognitive studies on these groups, already drew attention to their discovery that no significant differences were found between children with AD and children with PDD-NOS on any of the neuropsychological or behavioural measures, at least when non-verbal IQ was taken into account. However, due to the fact that the diagnosis of PDD is based on a smaller amount of autistic symptoms one might predict small differences between AD and PDD.

In conclusion, the present study found evidence for a deficient global processing in children with PDD, as well as a tendency for a more detail-oriented cognitive style as has been described for the group with AD. Taken collectively, the results suggest that perceptual problems that lead to weak coherence and that cause part of the executive dysfunctions might be an integral part of the deficiencies in the cognitive functioning of all children with PDD. Apart from this distinct cognitive style, executive dysfunctions might exist independently, as Booth et al. (2003) suggested. Hopefully, the present results will encourage more extensive research into information processing in the large group of children diagnosed with lesser variants of pervasive developmental disorders.

Acknowledgments

Wim A.J.M. Schlooz was funded by the Foundation for Child and Adolescent Psychiatry East Netherlands (SKJPON, Stichting Kinder- en Jeugdpsychiatrie Oost Nederland). The authors would like to thank Chantal Lankveld and Christine Trimbos for their assistance in testing the children. And we, of course, also thank all the children, their parents and schools for their invaluable help.

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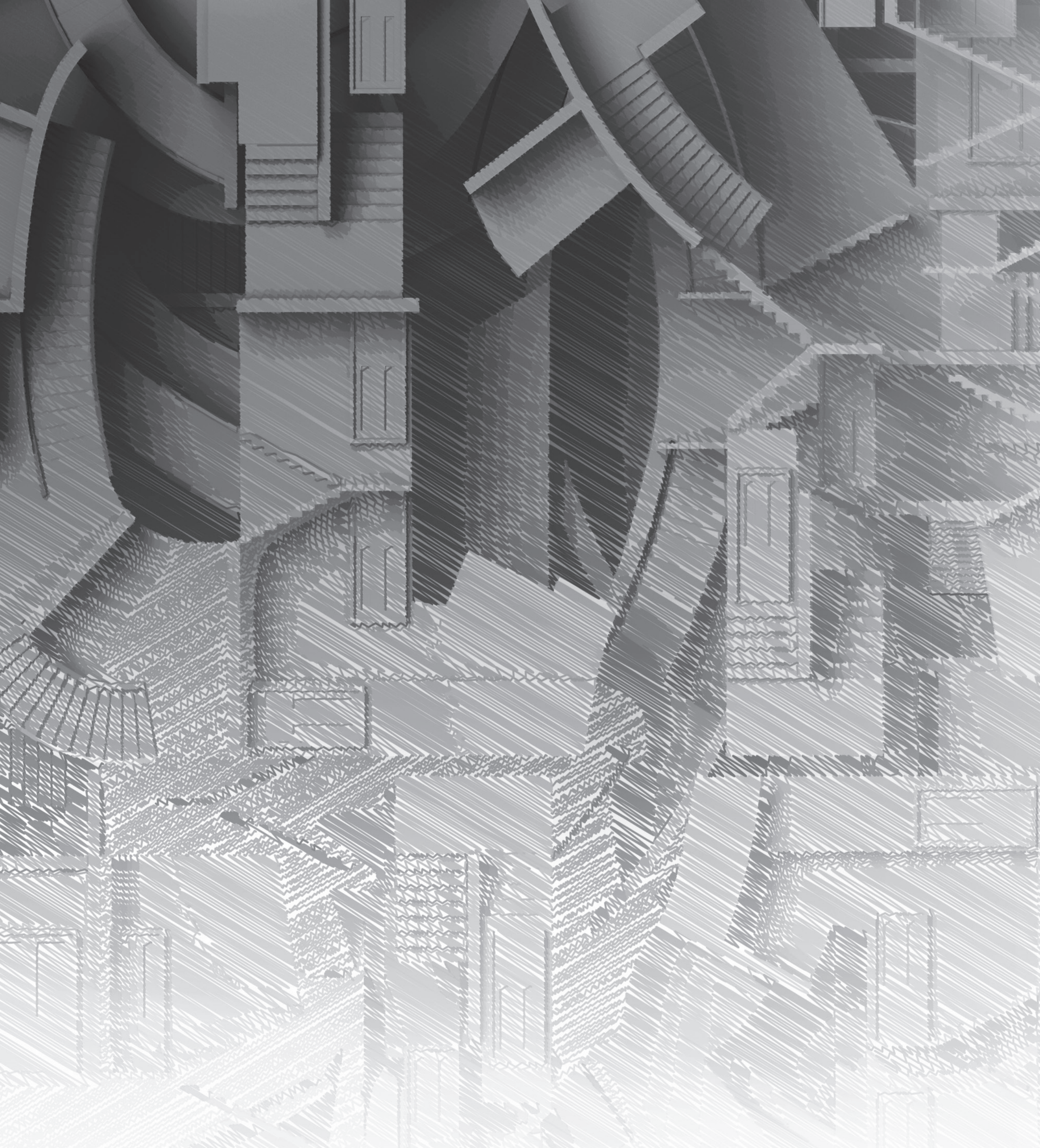

3

Atypical visuomotor performance in children with PDD

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Research in Autism Spectrum Disorders
(2012), 6, 326-336

ABSTRACT

Children with autism spectrum disorders (ASD) frequently encounter difficulties in visuomotor tasks, which are possibly caused by atypical visuoperceptual processing. This was tested in children (aged 9–12 years) with pervasive developmental disorder (PDD; including PDD-NOS and Asperger syndrome), and two same-age control groups (Tourette syndrome and typical developers) using two tasks: a visual and non-visual tactile tracking task (modified from Hermelin & O'Connor 1970 task) and the Developmental Test of Visual-Motor Integration (VMI). Both tasks revealed marked differences between the PDD group and the controls. Confirming Hermelin and O'Connor's findings in 'classical' autism, the children with PDD were faster than the controls on the non-visual tracking task, whereas they performed similarly to the controls when they could see the tracks. However, VMI copy scores were lowest for the children with PDD, while their scores on the visual perception and motor coordination subtests did not differ from the controls. The results support observations of an atypical visuomotor performance in children with PDD, which appears to derive from a deviant use of visual information in planning and guiding movements.

INTRODUCTION

Children with autism process and utilise visually presented information in an atypical, sometimes deficient, way. Evidence for this atypical visuoperceptual processing has been found in many studies. The performance of children with autism was found to be superior on Embedded Figure Tasks (Shah & Frith, 1983) and the Block Design test, while they did not succumb to visual illusions (Happé, 1996; Ropar & Mitchell, 1999, 2001). Also, tasks focusing on precategorical perceptual processing yielded superior performance outcomes for groups with autism: Navon-type tasks (Mottron, Burack, Iarocci, Belleville, & Enns, 2003), tasks involving disembedding a letter within a task-irrelevant context (Mottron et al., 2003), and visual search tasks (O'Riordan, Plaisted, Driver, & Baron-Cohen, 2001). For a recent and exhaustive review see Simmons et al. (2009). In children with a lesser variant of autism we found comparable results using the Rey Complex Figure task (Schlooz et al., 2006) and a visual completion task (de Wit, Schlooz, Hulstijn, & van Lier, 2007), indicating that also in this group there is a deviance in visuoperceptual processing, both at high and low levels of visual perception.

The question remains whether and how these atypical qualities in perception affect visuomotor performance. In clinical practice it is well recognized that children with ASD have great difficulties with tasks in which perceptual information has to be translated into motor activity. Sometimes, they simply seem unable to jump over a puddle, have serious problems catching a ball, riding a bicycle, or writing. This deviant visuomotor performance can either be explained by assuming that these children have additional (co-morbid) deficiencies in motor control (clumsiness), or by supposing that their atypical perception affects their performance of perceptual motor tasks.

Motor functioning deficits in autism have been widely studied (Ghaziuddin & Butler, 1998; Green et al., 2002; Manjiviona & Prior, 1995; Miyahara et al., 1997; Rinehart, Bradshaw, Brereton, & Tonge, 2001; Rinehart et al., 2006; Sturm, Fernell, & Gillberg, 2004). In the present study, however, we specifically examine the effects of visual perception on motor performance. Appropriate visual perception is not only required for an adequate identification of the object that is to be manipulated in perceptual motor tasks (goal selection), it is also a prerequisite for the translation of spatial locations into egocentric space (perceptual-motor integration; Willingham, 1998), for planning the movement trajectory, and for the guidance and control of the movement during its execution. According to the classic distinction proposed by Goodale and Milner (1992) and Milner and Goodale (2008), visual perception follows two paths in our brain, the 'what' and the 'how' route, the first supporting a proper identification of an object, and the other aiding the navigation of actions in the manipulation of this object. Most studies on visual perception in autism only addressed object perception, while the effect of vision on motor action has received far less attention. We know of no study that investigated this latter aspect in children with lesser variants of autism.

In collaboration with Frith, Hermelin and O'Connor (1970) analysed the effects of visual perception on the motor performance of autistic children using a groove tracking task. In their sensory integration study, children with autistic disorder (AD) and matched controls had to guide a metal stylus through a grooved track of a Perspex mould in two experimental conditions: one in which the children had full view of the track, of their arm and of their moving hand, and one in which the view of track and hand was blocked. In the 'no-vision' condition the stylus was guided by the grooves, affording the children tactile information only. In the 'vision' condition the children were able to see the entire track, to visually plan the route to the endpoint, and to monitor their motor actions. Because in this condition the stylus was also guided by the grooves, the children did not have to use the information they gleaned from their 'visual guidance' as Hermelin and O'Connor coined it. They could also, or solely, rely on the tactile input. In the original study, both groups were faster when they had full view of the tracks and their movements. But in the no-vision condition the control group was slower than the AD group. The authors suggested that children with AD might be less able to utilise vision for the planning of actions in perceptual motor tasks.

Several later studies also dealt with the effects of visual perception on motor performance in AD. Wing (1976) observed problems of motor imitation and Fulkerson and Freeman (1980) concluded that autistic children were deficient in monitoring their motor response in the drawing and copying tasks they used. In an experiment in which the effect of prism-induced visual displacement of objects was studied, Masterton and Biederman (1983) reported autistic children to rely on proprioceptive rather than visual feedback to modulate their motor output. Hughes (1996) suggested that impaired visual control of movement could account for the poor performance of the autistic children in her study on the execution of goal-directed motor acts. Gepner, Mestre, Masson, and de Schonen (1995) and Gepner and Mestre (2002) found no postural reaction to visually perceived motion information in AD. All these findings suggest that children with AD have problems deriving benefit from visual information during motor performance. Yet, contradictory findings were reported by Gidley Larson, Bastian, Donchin, Shadmehr, and Mostofsky (2008). In their carefully designed study in which children had to throw a ball at a target in a prism adaptation task, and move the handle of a robotic arm to guide a cursor to a target in a reaching task, they found no differences between autistic children and typically developing children in the adaptation of their motor output in response to a change in the environment.

In sum, the question of whether children with ASD have difficulties in availing themselves of visual information to plan and guide their movements is still open. To add to the debate, and using a digital recording technique to allow additional kinematic analyses, we decided to first try and replicate the tracking task data of Hermelin and O'Connor (1970), and to subsequently compare the outcomes on this task with scores obtained with the Beery–Buktenica Visual-Motor Integration test (VMI; Beery, 1967; revised 1997), a standard neuropsychological test designed to assess the extent to which children can integrate their visual and motor abilities. It requires the child to copy increasingly complex figures, and has subtests measuring the visual perception of these figures and the motor coordination required for drawing.

As alluded to above, in two earlier studies we also found that children with Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS) and Asperger syndrome (AS) had difficulties reproducing a complex geometric figure, and that they showed evidence of fragmented visual perception in a visual completion task (de Wit et al., 2007; Schlooz et al., 2006). In the current study we again only included children with these lesser types of autism.[1] Although together these two groups far outnumber the population of children with AD (42.9 versus 21.6 per 10,000 according to Fombonne, Zakarian, Bennett, Meng, & McLean-Heywood, 2006) and despite their constituting clinically important populations, they are understudied, and are generally diagnosed at an older age. They thus tend to go without adequate educational support for quite some time.

We compared primary-school-age children with PDD-NOS and Asperger syndrome to typically developing children (TD), and to an extra control group of children with Tourette syndrome (TS). Some authors (see Baron-Cohen & Joliffe, 1997) have postulated that ASD and TS share a number of similarities. Both are childhood developmental disorders, disrupting normal schooling and development of peer relations. Others claim that both disorders involve frontal abnormalities, share a genetically based aetiology, and affect boys more than girls (Pennington & Ozonoff, 1996). The inclusion of a TS group hence served to control for a-specific effects of a neuropsychiatric disorder. We, moreover, kept the window of chronological and mental age as small as possible to control for age-dependent effects: the

[1] Children with AD, Rett syndrome and childhood disintegrative disorder were excluded.

children were all functioning in a very comparable and relative stable stage of life, between young childhood and the start of adolescence. As the DSM IV does not provide explicit diagnostic criteria for PDD-NOS, Buitelaar and colleagues formulated diagnostic rules for this population (Buitelaar & van der Gaag, 1998; Buitelaar, Van der Gaag, Klin, & Volkmar, 1999). Their criteria and diagnostic rules proved to be more effective in segregating PDD-NOS from non-PDD cases than was possible on the basis of the diagnostic rules prescribed by the DSM IV. We adhered to these more narrowly defined diagnostic rules for PDD-NOS, and thus for the whole group diagnosed with PDD2 in the present study. Finally, none of the children participating in our trial used medication to preclude drug-induced effects.

We hypothesized similar performance results for our PDD sample on the tracking task to those (Hermelin & O'Connor, 1970) reported for their AD sample. We accordingly expected our children with PDD to perform faster in the no-vision condition than their age-matched controls. We also assumed them to score lower than the controls on the VMI, with the largest group differences becoming evident in the copying task, with no or smaller group differences in the visual perception and motor coordination subtests.

METHOD

Participants

A group of 12 boys with PDD aged between 9 and 12 years (see Table 3.1) were compared with an equal number of same-aged boys with Tourette's (TS) and typically developing boys

Table 3.1 Study group characteristics

Group (*n* = 12)	Chronological age	Full-scale[*] IQ	Verbal IQ	Performance IQ
PDD[#]				
Mean	10.5	105.8	105.3	103.9
Range	9.2–12.10	85–135	81–136	88–125
TS[+]				
Mean	11.3	106.7	105.3	107.0
Range	9.5–13.2	83–131	80–122	79–135
TD[~]				
Mean	10.6	Normal-range		
Range	9.5–11.5	School performance		

[a] The mental ages of the children with PDD and TS were established using the Wechsler Intelligence Scale for Children – Revised (WISC-RN). [b] PDD: PDD-NOS (*n* = 9) and Asperger syndrome (*n* = 3). [c] TS: Tourette syndrome. [d] TD: typically developing.

(TD). The clinical subjects were recruited from a regional outpatient clinic associated with the outpatient *Clinic of Child Behavioural Neurology* and the *Academic Centre for Child and Adolescent Psychiatry* of the *University Medical Centre St Radboud Nijmegen*. Their mental functioning was assessed with the Dutch Version of the Wechsler Intelligence Scale for Children – Revised (WISC-RN; Wechsler, 1974). The children in the TD group were recruited from a local primary school. None of the children used medication. Informed consent was appropriately obtained from all children and their parents.

The children with PDD had been examined by experienced clinicians, child psychiatrists and child psychologists both independently and together, and all fulfilled the relevant DSM-IV diagnostic criteria (APA, 1994). Applying the more restrictive scoring rules for PDD-NOS based on ICD 10/DSM IV criteria (Buitelaar & van der Gaag, 1998; Buitelaar et al., 1999) we found none of the boys to have AD, while three were diagnosed with Asperger syndrome and nine with PDD-NOS.

The selection procedure for the Tourette group was identical to that of the PDD group. A child neurologist, child psychiatrist, child neuropsychologist, and a physiotherapist, all part of the children's dedicated treatment team specialized in the treatment of childhood TS, based their diagnoses on the relevant DSM-IV criteria. All children were video-monitored during their visits to the physiotherapist. Children with comorbidity were excluded.

The typically developing boys were assessed by their own teachers and the psychologists who were to administer the experimental tasks. Selection was based on an evaluation of the children's school performance: rated on a 5-point scale, children with a score of either 1 or 5 were excluded from the study. All eligible children functioned at a normal cognitive level.

Table 3.1 lists the chronological ages for all three groups and the outcomes for the WISC-RN for the two clinical groups.

Material

Performance on all tasks was digitally recorded by means of a graphics tablet (WACOM 1218 RE) and two wireless electronic pens, with pen positions being sampled at a rate of 200 Hz, with a spatial resolution of 0.2 mm. The recorded tracking and drawing movements were analysed offline and split into movement times and stop intervals (OASIS; de Jong, Hulstijn, Kosterman, & Smits-Engelsman, 1996).

Measures

Tracking task

The tracking task partially repeated the Hermelin and O'Connor task (1970). In our task the children had to guide a non-inking stylus along a 5 mm deep and 2.5 mm wide groove in a Perspex sheet placed on the digitizer. As in the original study, we presented four tracks (and an additional practice track) of equal length, consisting of 24 segments and one track of 25 segments. Note that we used 2-cm long segments rather than 1-in. ones to match the size (A3) of the digitizer. The first track was a straight line and served as a practice trial only, while the four experimental tracks were of increasing complexity (see Figure 3.1). In the vision condition the children had full view of the track and their movements, while in the no-vision condition their view of the tracks and their hands was blocked by a custom-made wooden cover.

Developmental test of visual-motor integration (VMI: Beery, 1967 revised 1997)

All children also completed the VMI as well as its two standardized subtests: the Visual Perception and the Motor Coordination tests. Participants are instructed to copy, compare (perception) and trace (motor coordination) a series of 27 geometric forms of increasing complexity. As the first three forms are specifically designed to assess children under the age of 8 years, we did not offer them to our groups; accordingly, form 4 became our form 1. Execution times were analysed in three complexity categories: the simple lines (numbers 1, 2, 5, and 7), the simple figures (numbers 3, 4, 6, 8, and 9), and the complex patterns (10–13, 15–17, and 19). We further slightly modified the presentation of the forms: instead of in a booklet we presented the various stimuli on separate sheets of paper, each of which could be placed on the digitizer.

Procedure

The two clinical groups were assessed in the Nijmegen clinic and the TD group at their own school. Three experimenters conducted the tests; working in pairs, one experimenter instructed and assisted the child, while the other operated the equipment and coordinated the recordings. Each child was tested individually and all children first completed the tracking task and then the VMI.

In the no-vision condition of the tracking task a wooden cover with a small curtain at the open end (see Figure 3.1) was placed over the tracks preventing the children from seeing

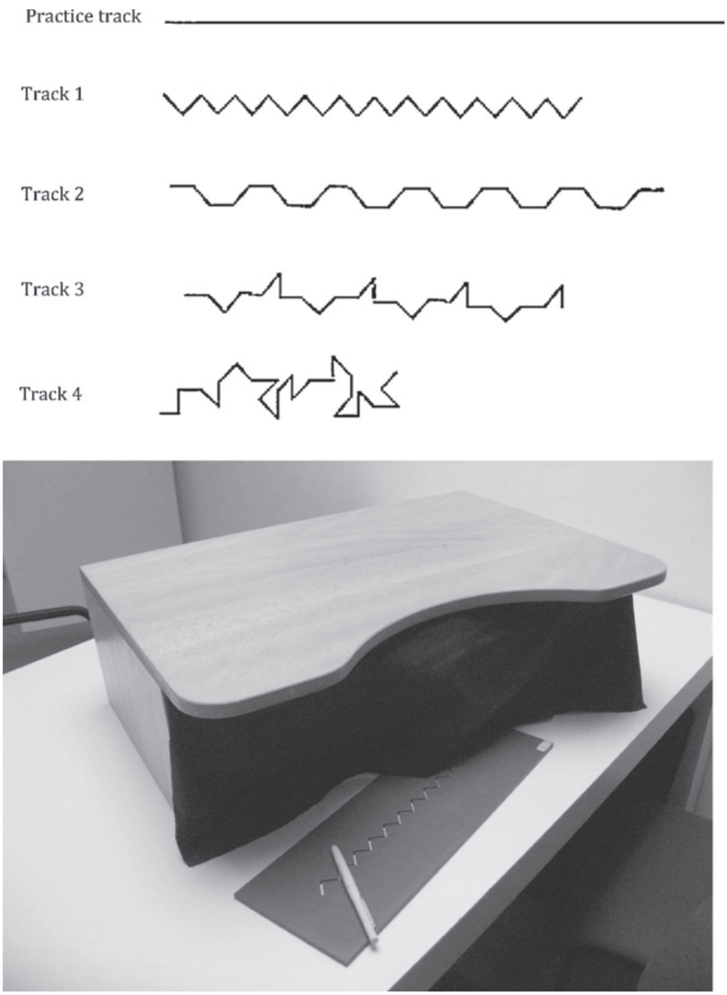

Figure 3.1 Tracking task. Representation of the four tracks to be traced in the tracking task and the set-up in the 'no-vision' condition of the tracking task.

the tracks and their active hand while still affording them ample room for manoeuvre. All children completed both conditions with their preferred hand, with the order of the vision and no-vision conditions being balanced. Verbal instructions were minimal ("Here you can see/Here you can feel a track. Please, place your pen here, inside the groove, and then move it to the end of the track as quickly as you can") and supplemented by a demonstration and a practice run. Execution time (ET) was measured per trial from the moment the pen left the starting point up to and including the moment the pen reached the endpoint of the track, thus covering the time during which the pen was moving, as well as the intervals during which the pen stopped moving. Offline, this overall ET was subdivided into a summed movement time and a summed stop time.

Implementation and scoring procedures for the VMI were in accordance with the standard instructions (Beery, 1997). All participants first performed the VMI (copying), then the visual-perception subtest and lastly the motor-coordination subtest. Scoring was to be stopped when the participant had proven unable to successfully draw three consecutive figures, but all children succeeded in reproducing all shapes.

Data analysis

Hypotheses were tested with one-tailed non-parametric statistical tests. The Kruskal–Wallis test was used to establish between-group differences per condition, the Mann–Whitney *U*-test to analyze the differences between the PDD group and each control group, and the Wilcoxon signed-ranks test for within-subject differences between two conditions. Statistical significance was set at $P < .05$.

RESULTS

Tracking task

Execution times

In Figure 3.2 the execution times are shown averaged over the four tracks for the PDD group and the combined control group. For comparison, the figure includes the original graph by Hermelin and O' Connor (1970, p. 56). In the vision condition the ETs did not differ between the PDD group and the controls (8.30 s versus 9.17 s; $U = 162$; $P = .27$), but in the no-vision condition the PDD group was fastest (23.7 s versus 16.3 s; $U = 95$; $P = .05$).

Figure 3.3 presents the mean ETs per track for the three groups separately. Because the outcomes of the TS and the TD group were all very much alike and because statistical tests did not reveal any significant differences, the two groups were pooled into one control group in the statistical analyses, which was then compared to the PDD group. None of the tracks showed a significant group difference in the vision condition, but in the no-vision condition the PDD group was faster on all tracks, with significant differences on tracks 2 and 3 (track 1: $U = 103$, $P = .09$; track 2: $U = 86$, $P = .026$; track 3: $U = 67$, $P = .005$; track 4: $U = 123$, $P = .24$).

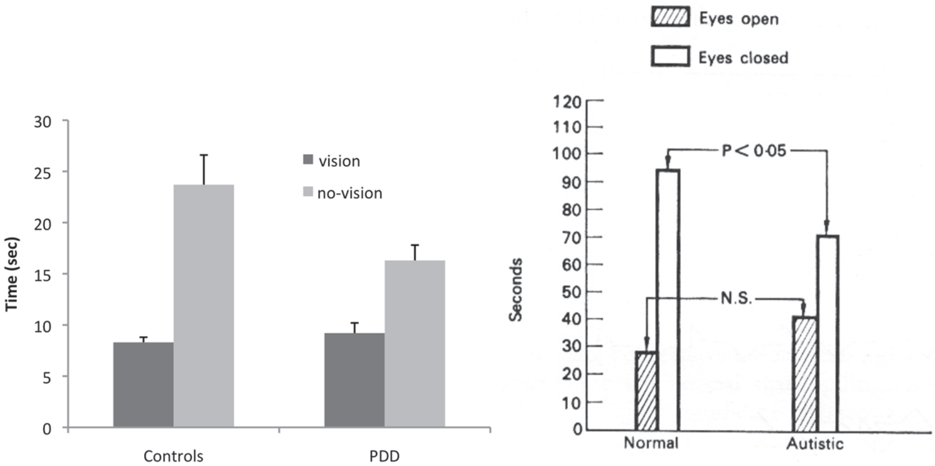

Figure 3.2 Execution times. Left-hand graph: execution times for the combined control group and the PDD group averaged over the four tracks of the tracking task per viewing condition. Right-hand graph: the original Hermelin and O'Connor figure (1970, p. 56) displaying the "average time (s) to complete 1 track".

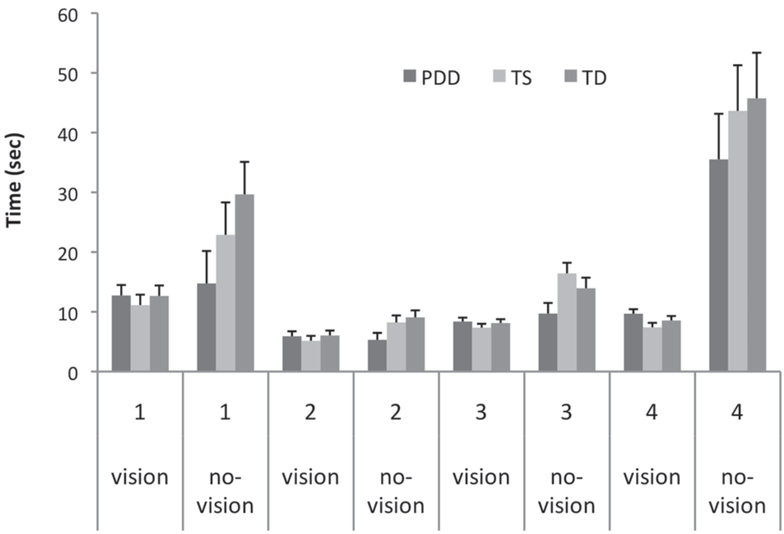

Figure 3.3 Execution times per track. Mean Execution Times (s) per track (1–4) and viewing condition of the tracking task for the three study groups. Error bars represent Standard Errors (SEs).

Figure 3.3 also illustrates that in the no-vision condition the performance on track 4 greatly differed from the performance on the other tracks. Of course, track 4 was the most complex pattern (see Figure 3.1) given its many acute twists and turns, its greater length (50 cm instead of 48 cm) and its high level of irregularity.

Effect of condition order

Tracks 1, 2 and 3. Subsequent analyses unexpectedly revealed that the order of the conditions (vision/no-vision) had a surprisingly large impact (see Figure 3.4). Here, we applied two-tailed tests, while the fourth track was analysed separately. As the bars on the left-hand side in Figure 3.4(a) show, the mean ETs on the first three tracks for the controls that started with the no-vision condition were much higher in the no-vision condition than they were

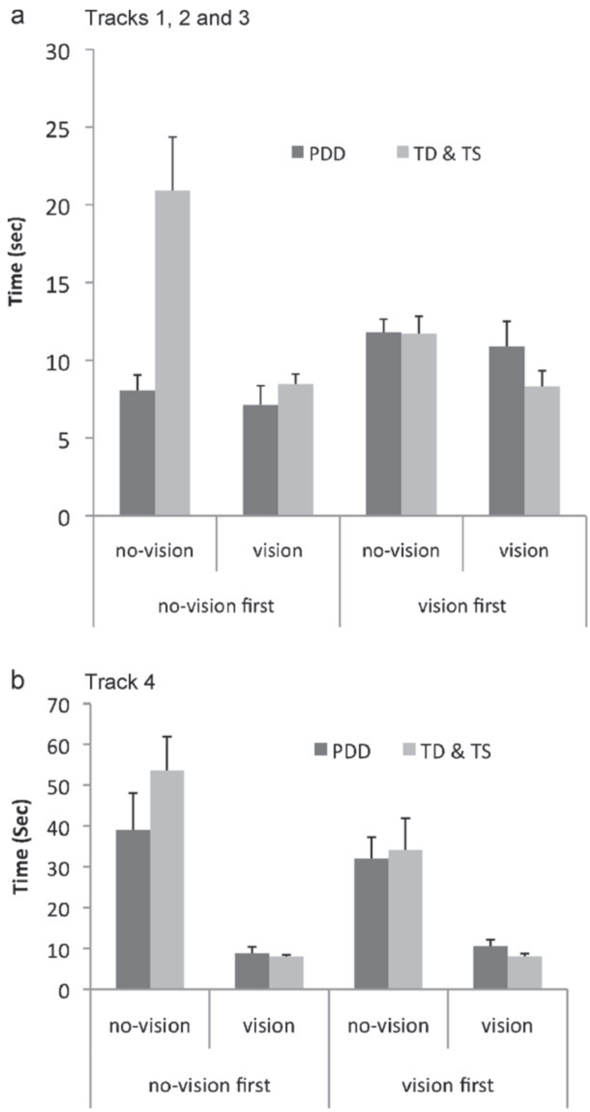

Figure 3.4 Execution times: condition order. The impact condition order of tracks 1, 2, and 3 (a) and track 4 (b) of the tracking task.

in the vision condition (21 s versus 9 s, $Z = 3.110$; $P = .002$). In contrast, the control children that had seen the tracks first were much faster in the no-vision condition and the difference they displayed between the two conditions was small (vision 12 s, no-vision 8 s; $Z = 2.312$; $P = .021$; see the right-hand side of Figure 3.4(a)). For the children with PDD the order of conditions had no effect. When the no-vision condition was presented first, their ETs were similar in both conditions (no-vision 8 s, vision 7 s; $Z = 0.734$; $P = .463$), as was the case when they were offered the vision condition first (no-vision 12 s, vision 11 s; $Z = 0.524$; $P = .600$).

Track 4. All children completed track 4 faster in the vision condition (see Figure 3.4(b)). The controls that had not seen the track before tended to be slower in the blinded condition than their peers who had performed the track with full vision first (54 s versus 34 s; $U = 39$; $P = .06$). For the PDD subgroup the effects of having seen the tracks were much smaller (39 s versus 32 s; $U = 16$; $P = .75$).

Execution times further explained. Splitting up ETs into movement and stop times revealed that in the no-vision condition the PDD group moved faster and significantly so in tracks 2 ($U = 85$; $P = .048$) and 3 ($U = 79$; $P = .029$). They also had shorter stop times, but again this was only the case in tracks 2 ($U = 86$; $P = .052$) and 3 ($U = 48$; $P = .001$). Analogous to the total ETs, the vision condition showed no significant between-group differences in movement and stop times.

Developmental test of visual-motor integration

Standard and subtest scores

The mean scores on the VMI and its two subtests are shown in Figure 3.5. The VMI standard scores displayed significant differences between the PDD and the two control groups ($\chi^2 = 9.01$; $P = .005$). The level of visual-motor integration was similar for the TD and the TS group ($U = 50$; $P = .10$), but the PDD group differed significantly from the *combined* control group ($U = 59.5$; $P = .003$). Contrary to expectations, neither the motor-coordination ($\chi^2 = 2.05$; $P = .18$) nor the visual-perception test ($\chi^2 = 4.98$; $P = .39$) revealed any such differences.

Execution times

There were no differences between the two control groups in the time they had taken to complete the patterns (lines: $U = 51$; $P = .22$; simple figures: $U = 43$; $P = .09$; complex figures: $U = 65$; $P = .68$). The children with PDD were faster than the combined control group on all three pattern categories: ($U = 63$, $P = .014$; $U = 67$, $P = .02$; and $U = 48$, $P = .004$, respectively).

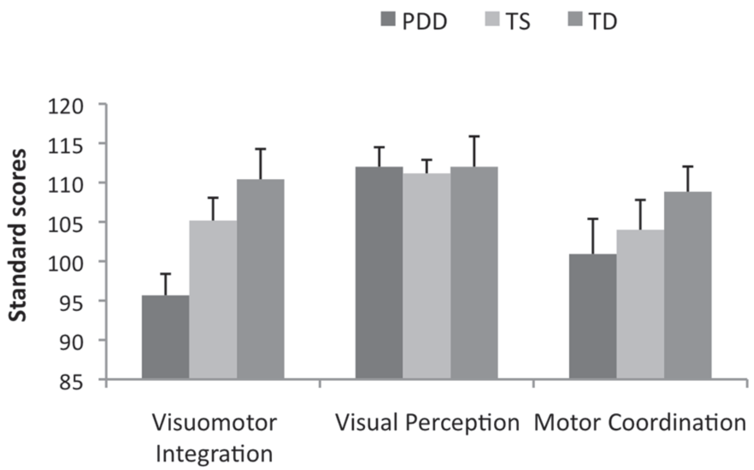

Figure 3.5 VMI standard scores. Scores on the VMI and its two subscales for the three study groups (*n* = 12).

DISCUSSION

With the present study we sought to explore how children with two lesser variants of autism, PDD-NOS and Asperger syndrome, make use of visual perception in motor planning. We compared their performance on a tactile tracking task with and without visual control and a standard test for visual-motor integration (VMI) with the outcomes obtained in two same-age control groups of typically developing boys and boys with Tourette syndrome. In both tasks the performance of the children with PDD clearly differed from that of the controls. In the tracking task all groups were equally fast in the condition in which they could see the tracks and monitor their movements. In the condition in which their view of the tracks was blocked, however, the boys with PDD performed faster than the controls. Without vision, the performance of the pooled control group deteriorated much more than that of the PDD group. These outcomes are in line with the results (Hermelin & O'Connor, 1970) obtained with their tracking task in children with autism. Whether they also support their conclusions will be discussed below. The VMI scores of the PDD boys were lower than those of the controls, as was expected. However, the VMI perception and motor coordination subtests did not show any between-group differences.

Our data also showed considerable differences with the results Hermelin and O'Connor reported. The execution times in our study were much faster (averaging 14 s) than the times they recorded in their experiment (about 58 s). This disparity may be attributable to the differences in the (perceptual) ages of the children tested (9–12 years in our trial versus 4–6 years in theirs), the track lengths (48/50 cm versus 24/25 in.), and small differences in materials. Moreover, our focus was on children with lesser variants of autism. Unfortunately, Hermelin and O'Connor did not report results for the four tracks separately. Our track analyses revealed marked differences between the first three tracks and the final fourth track. In tracks 1–3, the lack of vision in the blinded condition did not result in any performance decrement in the PDD group, but it did greatly slow down tracking times in the controls. Apparently, the children with PDD were superior here in exploiting tactile cues, while the controls relied more strongly on the visual information. Due to its non-repetitive character and its occasional directional changes, the course of the fourth track is unpredictable, requiring planning and control based on visual information rather than haptic guidance alone, even for the PDD group. Yet, they still showed a tendency to perform faster in the no-vision condition.

We also found marked effects of condition order. Again, the differences between the PPD group and the controls were considerable, especially in the first three tracks. The controls that first completed the vision condition appeared to use the information obtained during this condition for the subsequent no-vision trials, as was reflected by the reduced execution times on the covered tracks. The controls starting with the no-vision condition had no such advantage. Remarkably, in the PDD group condition order had no effect in the first three tracks. In track 4 the order effect was still smaller for the PDD group than it was for the controls. Overall, the controls seemed to profit more from having seen the tracks before than the PDD group.

We also looked at movement time and stop intervals (including pauses and pen-up times) separately. The supposition behind this distinction was that part of the duration of the stops might reflect the time needed to plan the subsequent movement. We found that the stop times were fewer and shorter in the boys with PDD, suggesting that they took less time to plan the next movement than the controls did. However, their summed movement times per track were also faster, which excludes a simple planning explanation. Given that they were also significantly faster on the VMI, our results reveal an overall speeding-up of both planning and motor execution in children with PDD.

With the performance differences between the vision and the no-vision condition being smaller in the autistic group, (Hermelin & O'Connor, 1970) suggested that children with AD benefit less than normally developing children from 'visual guidance'. However, it seems more appropriate to state that children with PDD considerably outperform their unaffected peers when no visual information is available. Seeing the tracks in the start condition only benefited the controls' performance in tracks 1, 2 and 3 in the subsequent blinded condition. Here, the controls seem to be impaired, not the boys with PDD. More specifically, having learned or being used to rely heavily on visual information – even when this information is dispensable because the available tactile information is sufficient to guide their actions – non-ASD children are more affected when it is not available than are children with ASD. Hermelin and O'Connor (1970) already postulated a higher proficiency in the use of tactile information in AD. The present results suggest that also children with lesser variants of autism are better able to solely use tactile and kinaesthetic information or have learned to take better advantage of this information.

The VMI scores of the PDD group were lower than those for the controls. But this was true for the standard (copying) test only; its two control tests failed to show any between-group differences. Copying entails a number of processes, among which the perception of the target shape, the construction of a representation, its storage in short-term memory, visual-spatial planning, and preparation of the most efficient sequence of movements, the execution of the motor commands and a monitoring and modification of the resulting graphic reproduction (see also Mottron, Belleville, & Ménard, 1999). Therefore, lower scores on the VMI copying test do not necessarily imply problems in visual-motor integration: they also support the notion that children with ASD process visual information atypically. The comparable group scores on the visual-perception task, however, suggest that our PDD children did not differ from the controls on this aspect. Yet, in this subtest children can easily compare two stimulus figures by simply matching only some of their aspects without perceiving the whole form. In the VMI copying test, on the other hand, not perceiving the whole figure is likely to hamper the construction of a correct representation in short-term memory, which will lead to inferior planning and hence an inferior reproduction of the original figure. In this vein, problems in forming an adequate representation in consequence of deficient visual processing might then explain the lower VMI scores in the PDD group, while the influence from visual information on motor activity might as such be unaffected. Also Klinger and Dawson (2001) pointed to the difficulties children with AD have in forming representations. This would be consistent with the results of an earlier study of ours in which we had children with PDD copy and reproduce the Rey Complex Figure (Schlooz et al., 2006), which pointed to visuoperceptual problems, leading to a more fragmented information processing hampering the reproduction of the complex figure. De Wit et al. (2007) also found precategorical visuoperceptual problems in PDD in their study on visual completion, in that already at a low level the formation of visual representations was disturbed.

The PDD group took less time to draw the VMI figures (correct trials only), irrespective of their complexity. They were even faster in drawing the simple lines (PDD: 3.8 cm/s; Controls: 2.9 cm/s), which, like in the tracking task, again suggests an overall speeding up of motor performance. Reduced visuoperceptual control and thus a reduced reliance on visual monitoring during drawing might account for this speeding-up phenomenon. More research is needed to confirm whether children with PDD are indeed faster because they spend less time planning and correcting their movements on the basis of visual feedforward and feedback.

Summarising, their faster performance on our two visuomotor tasks supports the idea that children with PDD rely less on visual information than controls do. In both the tracking task and the VMI they appear to gain time by using visual information to a lesser extent to plan and correct their movements, while their inferior scores on the VMI figure copying task underscore their reduced ability to integrate visual perception and motor execution.

Poor planning skills as part of executive dysfunction, could explain the lower VMI scores of the PDD group. Executive dysfunction is frequently seen in ASD (for a review, see Hill, 2004). Booth, Charlton, Hughes, and Happé (2003), Geurts et al. (2008), and more recently Bramham et al. (2009) found evidence of planning problems in ASD. Yet, planning presupposes there is something to be planned, and that, if the formation of a visual representation is impeded, planning will be impeded too. The results of our study do not allow the conclusion that we are dealing with poor planning resulting from visuoperceptual problems or with deficits deriving from an overall executive dysfunction. Moreover, a problem in planning, or an overall executive dysfunction, does not explain the superior performance of the PDD group on the tactile tracking task. Difficulties forming an abstract representation of the visual environment, which leads to a diminished use of visuoperceptual information and a greater reliance on tactile and kinaesthetic information, as well as a tendency for an overall speeding up of motor activities, however, does. This account would explain the performance differences in the tracking task and the VMI. The results in this study are hence more in line with the weak coherence account as formulated by Frith (1989): there seems to be a diminished use of visual information for motor action in ASD. Another explanation is that children with PDD may have problems in constructing the allocentric space, and/or in translating it into the egocentric space, or in constructing an egocentric space. Such deficiencies would render them less able to properly attune their inner world to the outer world, accounting for their problems with catching a ball, writing, and all other situations in which perception is needed to control the motor action.

A final note on the results for the boys with Tourette's. On both tasks, their performance was in the same range as that of their typically developing counterparts. Here it is important to reiterate that in our study none of the children with TS had any comorbidity and none used medication. Our results consequently indicate that in TS inferior performance on VMI-like tasks, measuring executive functions, is far more likely to be associated with comorbidity and not with TS per se, as Verté, Geurts, Roeyers, Oosterlaan, and Sergeant (2005) concluded earlier.

CONCLUSIONS

In the present study of school-age boys with PDD-NOS and Asperger syndrome, we found convincing evidence that in these lesser types of autism the quality of performance is similar to the performance found in children with full-blown autism. Our all-male sample clearly displayed a deviant perceptual-motor performance, which has serious implications for many activities of daily living, such as getting dressed, writing, and cycling. To support these children more effectively, we need to gain better knowledge of the nature of this anomaly. For instance, it is as yet unclear whether their perceptual-motor limitations mainly arise from perceptual problems (a reduced ability in forming a global representation that can guide actions) and/or from deviances in integrating perceptual information with motor planning and execution. Disentangling visual-perceptual processing from motor execution might lead to an answer. As tactile processing seems be enhanced in PDD, it may also be worthwhile to examine whether this dexterity can be exploited when learning or perfecting daily-life skills. Benefits are, however, likely to be restricted to the less complex skills that do not require the use of higher-order, visual-cognitive representations.

Acknowledgements

Wim A.J.M. Schlooz was funded by the Foundation for Child and Adolescent Psychiatry East Netherlands (Karakter) and Reinier van Arkelgroep. The authors would like to thank Christine Trimbos, Pieter van den Broek and Chantal Lankveld for their assistance in testing the children. And we, of course, also thank all the children, their parents and schools for their invaluable help.

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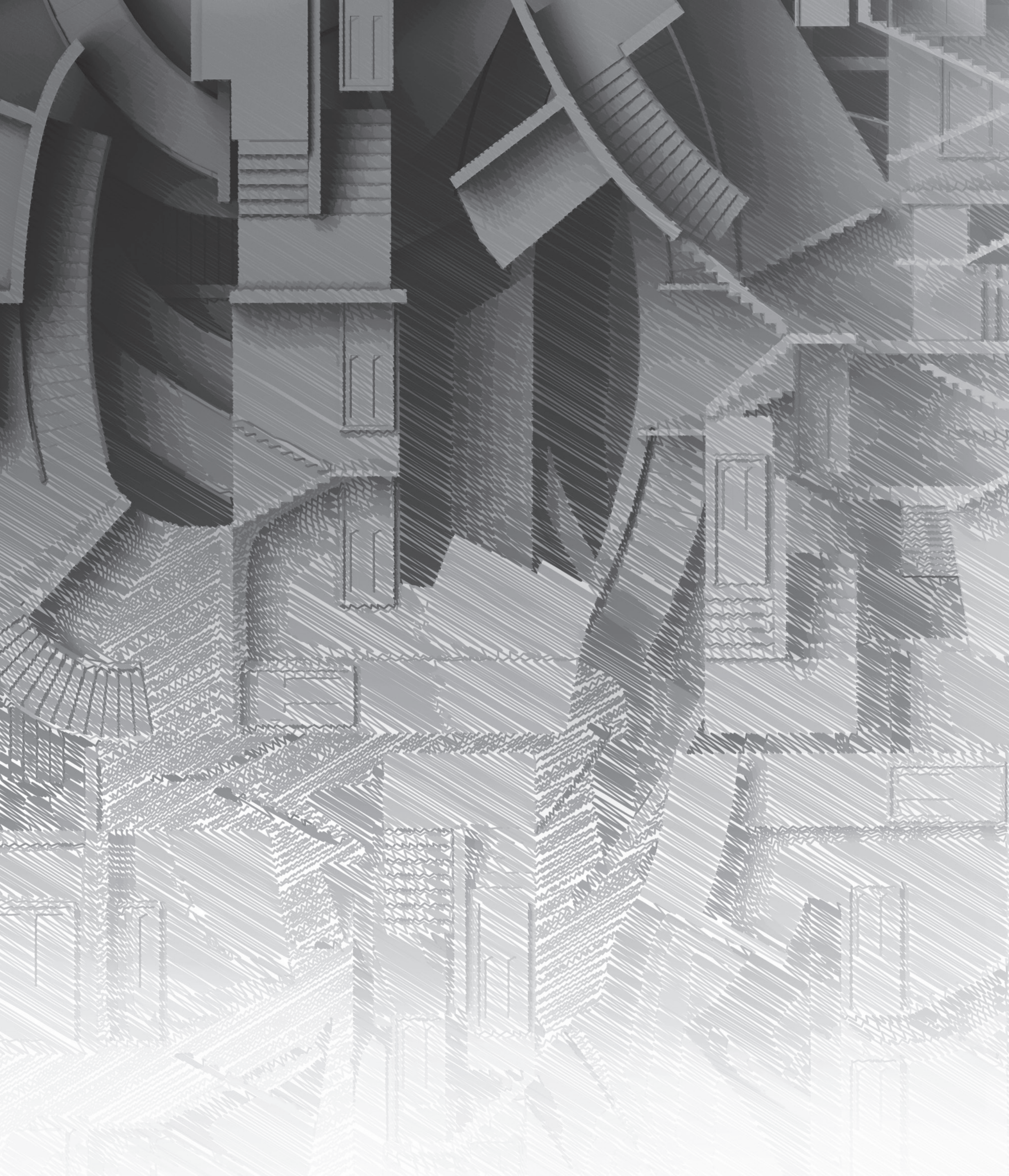

4

Boys with autism spectrum disorders show superior performance on the adult Embedded Figures Test

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Research in Autism Spectrum Disorders
(2014), 8, 1-7

ABSTRACT

Weak central coherence is frequently studied using the Embedded Figures Test (EFT) yielding mixed and ambiguous results. In this study, the performance of 36 boys (9–14 years) with Autism Spectrum Disorders (ASD) is compared with that of 46 typical peers using both the children's and the adult version of the EFT. Only in the adult version did the ASD group outperform the controls in terms of accuracy. Corrected for age and pIQ, a subgroup of boys with Autistic Disorder (AD) showed superior perceptual processing capacities, while the performance of boys with PDD-NOS and Asperger Syndrome was in between that of those with AD and the controls. The findings strongly suggest that children and adolescents with ASD will only show superior results on visual-perceptual tests if the task complexity and thus their sensitivity is sufficiently high to challenge typically developing age-matched peers.

INTRODUCTION

Typically developing and mentally handicapped children process meaningful and patterned information better than they do random and meaningless stimuli. People with autism profit less from meaning or Gestalt, but on the other hand show a remarkable ability to detect a target in visual search tasks (Almeida, Dickinson, Maybery, Badcock, & Badcock, 2010a; Almeida, Dickinson, Maybery, Badcock, & Badcock, 2010b; Jarrold, Gilchrist, & Bender, 2005; Kaldy, Kraper, Carter, & Blaser, 2011; O'Riordan, Plaisted, Driver, & Baron-Cohen, 2001; O'Riordan, 2004; O'Riordan & Plaisted, 2001; Plaisted, O'Riordan, & Baron-Cohen, 1998). In their now classic research note on autism, Shah and Frith (1983) were the first to show that children with autism possess a striking ability to detect hidden, embedded elements in a larger complex figure.

Yet, the experimental evidence of this salient ability is mixed. In their extensive review of studies on vision in Autism Spectrum Disorders (ASD), Simmons et al. (2009) discussed all studies published at the time in which either the children's or the adult version of the Embedded Figures Task (EFT) or both versions were used. They judged the balance of evidence to favor a superior performance by people on the autism spectrum. More recently, however, White and Saldaña (2011) reviewed another 16 studies exploiting the EFT and concluded that findings were inconsistent. Moreover, in their own study featuring two large samples of high-functioning children with ASD, they found their two groups to perform similarly to the groups of typically developing (TD) children. A closer examination of the White and Saldaña (2011) review learns that of the 14 studies of which the accuracy scores were listed, 11 studies showed that the percentage of correctly executed trials for the TD participants exceeded 75%; in five studies the accuracy rate even exceeded the 85% mark. Such high scores for controls easily lead to ceiling effects, making it more difficult to detect a statistically significant superior performance in the clinical group. In children with ASD superior visual–perceptual abilities might then only become manifest if the difficulty of the task is sufficiently high to challenge the controls (see Schlooz et al., 2006).

The present study was set up to test this idea. To try and avoid ceiling effects as much as possible, we presented a boys-only clinical and control group with the most complex figures of the Children's EFT only. In addition to these still relatively easy tasks, we also had both groups complete the adult version of the EFT, whose figures and forms are more abstract than those of the CEFT (see Figure 4.1), rendering the task more difficult, which, we assumed, was more likely to prevent ceiling effects from occurring.

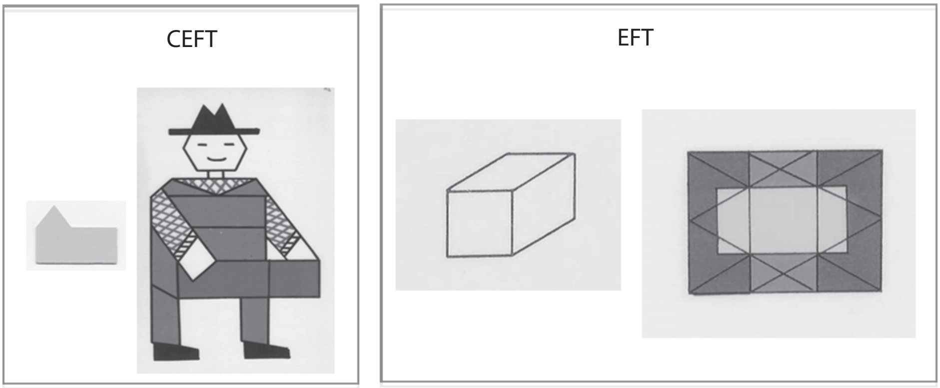

Figure 4.1 Embedded Figures Tests. Examples of the stimulus cards depicting one of the simple geometric forms (depicted left in each panel) to be identified and traced in one of the complex figures of the children's (CEFT – left panel) and the adult version of the test (EFT – right panel).

In the instructions about the norm scores of the CEFT Witkin and colleagues (Witkin, Oltman, Raskin, & Karp, 1971) point to the strong effect of age on its performance results. To control for any such age-dependent effects, we kept the window of chronological and mental age as small as possible. In our analyses of our clinical group of high-functioning boys with ASD, we furthermore looked separately at the boys diagnosed with Autistic Disorder (AD) and a subgroup consisting of boys with milder ASD, i.e. Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS) and Asperger Syndrome (AS), which will be referred to as the Pervasive Developmental Disorder (PDD) group. We made this distinction between AD and PDD firstly to be able to compare the results with those reported in the original study of Shah and Frith (1983), in which the clinical group consisted of children with autism only. Secondly, we hoped the distinction would also help us identify potential performance differences for the boys with diagnoses at the mild and more severe end of the autism spectrum. And finally, the distinction allowed us to examine the effects of age and intelligence on EFT performance in greater detail.

We expected the boys with ASD to perform similarly to their typically developing peers on the CEFT but to show superior performance on its adult version. We further expected the AD group to reach higher scores on the EFT than the PDD group.

METHOD

Participants

For reasons of availability and comparativeness, the children we recruited for our trial were all male and in a comparable stage of life, i.e. between young childhood and the start of adolescence (9–14 years) and functioning as can be expected at that age or given their diagnosis. The boys with ASD were recruited from the outpatient Clinic of Child Behavioral Neurology and the Academic Center for Child and Adolescent Psychiatry of the University Medical Center Nijmegen and an associated outpatient child psychiatric clinic. The controls, all typically developing boys, were recruited from a local primary school. Exclusion criteria were the use of medication and abnormal mental functioning. In all children mental functioning was assessed with the Dutch Version of the Wechsler Intelligence Scale for Children – Revised (WISC-RN; Wechsler, 1974), with children with a pIQ below 75 being excluded. Informed consent was appropriately obtained, with all children and their parents consenting to participate in the study.

Buitelaar and colleagues (Buitelaar & van der Gaag, 1998; Buitelaar, van der Gaag, Klin, & Volkmar, 1999) defined limiting scoring rules for PDD-NOS based on ICD10/DSM-IV criteria. We adhered to these rules when selecting our clinical group. Experienced clinicians, child psychiatrists and child psychologists examined eligible children with ASD both independently and together, resulting in 36 participants fulfilling the DSM-IV diagnostic criteria for ASD (APA, 1994). Fifteen boys were diagnosed with AD and 21 with PDD, of whom five were diagnosed with Asperger Syndrome and 16 with PDD-NOS. The controls (n = 46) were screened with the Child Behavior Checklist (CBCL) and the Teacher Report Form (TRF; Achenbach, 1991). Boys with a clinical score on any of the subscales were excluded from the study. The chronological ages for all three groups and the outcomes on the WISC-RN (Wechsler, 1974) are listed in Table 4.1.

Material

Performance was digitally recorded using a laptop computer, a digitizing tablet (WACOM 1218 RE) and a wireless electronic non-inking pen, with pen positions being sampled at a rate of 200 Hz at a spatial resolution of 0.2 mm. The recorded movements and reaction times were analyzed offline by mean of custom-made software (OASIS; De Jong, Hulstijn, Kosterman, & Smits-Engelsman, 1996).

Table 4.1 The chronological ages for all study groups and their performance scores on the WISC-RN

Group	*N*	Chronological age (months)	Full-scale IQ (fIQ)	Verbal IQ (vIQ)	Performance IQ (pIQ)
ASD					
Mean	36	130.69	102.67	100.61	104.64
Standard deviation		15.30	14.90	17.82	14.89
Range		108–173	66–143	55–152	79–137
Autism					
Mean	15	131.27	93.20	91.07	97.47
Standard deviation		13.59	13.38	17.31	11.55
Range		111–157	66–118	55–122	79–114
PDD					
Mean	21	130.29	109.43	107.43	109.76
Standard deviation		16.73	12.16	15.14	15.11
Range		108–173	88–143	88–152	79–137
Controls					
Mean	46	130.11	108.43	107.22	107.46
Standard deviation		12.12	13.30	12.45	13.97
Range		113–153	81–135	82–132	75–138

Measure

Children's Embedded Figures Test and Embedded Figures Test (Witkin et al., 1971)

In both editions of the EFT participants have to find a simple, geometric form that is embedded in a more complex figure (see Figure 4.1). Having detected the embedded shape, the boys in our study were instructed to trace its outlines within the complex figure using a non-inking electronic pen as quickly and as accurately as possible. In a single session, the children all completed the six most complex figures of the CEFT (i.e., numbers 14–19, all with a 'house' form as the embedded picture), as well as the standard adult EFT (Form A) consisting of 12 colored complex figures and 8 simple forms. In both tests, the pairs of a target form and complex figure were presented on separate sheets of plastic-coated paper (A4 format) to prevent traces. Thus, in accordance with the procedure used by Shah and Frith (1983) in their study, the target forms remained available during task performance, preventing the children from having to work from memory.

Procedure

The sheets with the figure pairs were consistently positioned at a fixed place on the digitizing tablet and presented covered with a red piece of paper. Before the start of each trial the children needed to place the pen within a fixed area indicated on the tablet. After an auditory signal (three beeps) from the laptop, the red cover sheet was removed and recording started. Prior to the CEFT and EFT, detecting and tracing embedded forms was trained using discrete practice shapes and figures not used in the test trials. The children were instructed to try and find the embedded forms as quickly as possible and to subsequently trace them within the complex figures as quickly and as accurately as possible with the pen.

The dependent variables were accuracy defined as the percentage of correct responses given within the allotted time, which was set at three minutes, and mean reaction time. To enable us to compare the CEFT and EFT scores, we used the same scoring principle for both tests, which entailed a deviation from the procedure Witkin et al. (1971) prescribed for the EFT in that we did not correct the children when they made a mistake by choosing the wrong form within the complex figure. These attempts were simply noted as incorrect responses.

In Shah and Frith's landmark study (1983) the autistic children attained better accuracy scores and also tended to spot the embedded figures more quickly than the controls did. In the present study the mean reactions times were only used to determine the strategy the children adopted and to thus pinpoint whether any superior accuracy was achieved at the cost of longer response times or whether the higher percentage correct responses was reached in the same or even less time. Reaction time was measured from the time the red cover paper was removed until the moment the pen was displaced to trace the correct embedded form within the complex figure. If at first a wrong form was traced, which action was subsequently aborted and the correct form traced, then the start of the second tracing action was adopted as the reaction time. Only reaction times to correct trials were entered into the analyses.

To compare the study groups appropriately, matching was based on Performance IQ (pIQ) as children with autism generally have a lower Verbal IQ (vIQ) (Mayes & Calhoun, 2003; Shah & Frith, 1983). If matching is done using Full scale IQ (fIQ) scores, this might give children with ASD an advantage over typical age peers.

Data analysis

Pearson's correlations were used to analyze the relationships between group characteristics and CEFT outcomes. Differences between the full clinical group and the control group were analyzed with Student's *t*-tests (one-tailed probabilities are reported). Subsequently, also the AD and PDD subgroups were further analyzed separately by means of a GLM Univariate

Analysis of Variance (ANOVA) procedure with Group as the between-subjects factor. In all analyses *P*-values lower than .05 were considered to be significant.

RESULTS

CEFT and EFT performance scores

The means and standard deviations (SDs) of the main outcomes for the two tests are presented in Table 4.2. Confirming our hypothesis, the results on the CEFT yielded no significant group differences, while on the EFT the boys with ASD scored significantly better than the controls with respect to accuracy.

Covariation with age and IQ

In general, CEFT and EFT scores are age- and IQ-dependent (Bölte, Holtmann, Poustka, Scheurich, & Schmidt, 2007; Witkin et al., 1971). In the present study we found significant correlations between these variables and the accuracy rates for both tests. For the CEFT, the correlation between percentage correct and age was $r = 0.38$ ($P = .001$), while pIQ yielded a correlation of $r = 0.36$ ($P = .001$). For the EFT, correlations were $r = 0.42$ ($P < .0001$) for age and $r = 0.48$ ($P < .0001$) with pIQ. As to response times, correlations with age were significant in the CEFT ($r = 0.26$, $P = .018$) only, while correlations with pIQ were not significant for either test.

Because of the correlations obtained, age and pIQ were entered as covariates into the univariate analyses of variance that were performed to test the differences between the clinical and control groups. As was to be expected from the reported correlations, the two covariates produced significant ($P < .001$) results in the CEFT and EFT analyses of percentage correct and of CEFT reaction time. Figure 4.2 shows the mean results for the two study groups.

Table 4.2 Means (standard deviations) for the two tests and t-test results of the differences between the ASD group and the typical developers (control group)

	Children's Embedded Figures Test				Adult Embedded Figures Test			
	ASD	Controls	*t*	*P*	ASD	Controls	*t*	*P*
Accuracy (% correct)	91 (15)	87 (18)	0.85	.198	56 (24)	48 (22)	1.72	.045*
Reaction time (s)	13.2 (6.7)	11.7 (6.5)	1.04	.150	26.0 (13.0)	26.2 (11.9)	-.06	.476

* $P < .05$.

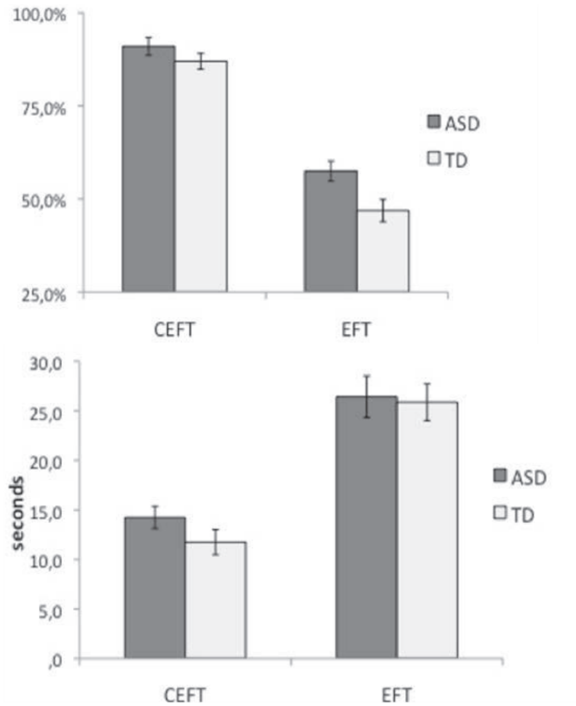

Figure 4.2 Accuracy (% correct) and reaction times. Scores corrected for age and pIQ, for the children's and adult Embedded Figures Test (CEFT and EFT, respectively) for the ASD and the typically developing (TD) controls.

CEFT performance

Performance of the ASD and TD groups was not significantly different on the CEFT neither in accuracy nor reaction times (percentage correct: $F(1,78) = 1.254$, $P = .218$; reaction time: $F(1,78) = 2.16$, $P = .145$).

EFT performance

In the adult version of the EFT the ASD-TD performance differed significantly on the accuracy measure (percentage correct: $F(1,78) = 6.97$, $P = .001$). Reaction times in the EFT showed the same picture as that recorded for the CEFT (see Figure 4.2) and there were no group differences ($F(1,75) = .039$, $P = .845$).

Three-group analysis

In a three-group (AD, PDD and TD) analysis with age and pIQ as covariates only the EFT accuracy produced significant results ($F(2,77) = 3.76$, $P = .028$). Table 4.3 presents the mean values and shows that the highest score on EFT accuracy was attained by the AD group with

Table 4.3 Means (standard errors) for the two versions of the visual-perception test corrected for age and pIQ for the two ASD subgroups, and the control group

	Children's Embedded Figures Test			Adult Embedded Figures Test		
	Autism	PDD	TD	Autism	PDD	TD
Accuracy (% correct)	88.2 (3.9)	93.0 (3.2)	87.0 (2.2)	60.4 (4.8)	55.5 (4.0)	46.8 (2.7)
Reaction time (s)	11.0 (1.7)	14.6 (1.4)	11.8 (0.9)	25.0 (3.4)	27.3 (2.7)	25.9 (1.9)

the PDD group achieving a mid-position. Simple contrasts on this variable revealed that the AD group scored significantly ($P = .017$) better than the TD group, while the percentage of correct responses for the boys with PDD ($P = .072$) was not significantly higher.

DISCUSSION

In this study we tested whether high-functioning boys (aged 9–14) with autism spectrum disorders would outperform typical age peers on the children's or the adult version of the Embedded Figures Test, as this latter version is more difficult than its adaptation for children that is generally used in childhood autism studies. Even though we only used its most complex items, the CEFT yielded no performance differences between the two groups, confirming our assumption. Consistent with our second hypothesis, on the adult version of the test, the boys with ASD performed significantly more accurately than their typical peers. Their superior performance was even more pronounced when the covariates age and pIQ were taken into account. Looking at the two clinical subgroups separately, we found the accuracy scores the boys with AD attained on the EFT to be significantly higher than those attained by the controls, while the scores for the boys with PDD were in between those of the boys with AD and those of the controls.

With accuracy scores on the CEFT (ASD: 91%; TD: 87%) being substantially higher than those for the EFT (56% and 48%, respectively), the CEFT was indeed much easier to complete successfully than its adult counterpart. Reaction times for the CEFT were also about half those recorded for the EFT. As hypothesized, to demonstrate the superior visual-perceptual performance of children with ASD on Embedded Figures Tests, the level of difficulty needs to be such that the controls are truly challenged.

Performance differences were manifest in accuracy scores only, not in reaction times. In interpreting the results, it must be remembered that, as explained in the Method section, our EFT task procedure deviated from the procedure Witkin et al. (1971) prescribed for the test. The children in our study were not corrected when they made a mistake in choosing the wrong form in the complex figure. In addition, we analyzed reaction times for the correct responses only to allow us to determine which strategy the boys had adopted when making their choice. Our reaction time results confirm that the superior accuracy in spotting the embedded shapes is not achieved by a different temporal strategy; the boys with ASD simply made fewer mistakes.

The moderately strong correlations we obtained between accuracy, and age and pIQ are in agreement with earlier findings. Witkin and co-authors (1971) already pointed to the strong age effects on the Embedded Figures Tests, while much later Bölte et al. (2007) mention a strong correlation with pIQ.

But how can our moderate but positive results be explained in the light of the many negative results White and Saldaña (2011) reported for previous studies? As mentioned earlier (Schlooz et al., 2006), an important factor in evaluating the results of studies using an EFT is the difficulty of the task as it affects its sensitivity; the task must be complex enough to prevent a ceiling effect. Also, ASD severity will influence the magnitude of the performance difference between the clinical and control groups. Thus, many of the studies White and Saldaña reviewed used the relatively easier CEFT, while they also included participants who were chronologically older and functionally at a relatively high mental level. None of these studies showed a superior performance for the clinical groups. However, when participants were younger and functioning at a lower mental age, the CEFT did reveal group differences. In their 2005 study, Jarrold, Gilchrist, and Bender, for instance, reported a much better performance for the children with ASD (mean age 15 yrs; Raven 52) than for the control group. van Lang, Bouma, Sytema, Kraijer, and Minderaa (2006) also found their ASD group to perform better on the more difficult items of the CEFT (accuracy ASD group 39%; controls 25%). Their participants were of adolescent age (mean 14.5 yrs), functioning at a mentally retarded level (pIQ 57) and very well matched with the control group. In Shah and Frith's 1983 study, the two control groups were matched for IQ or age with the autism group (Raven 75 and 72 respectively), and their accuracy was well below the ceiling (63%), leaving room for the children with autism to score better (82%).

Also in studies that use the adult version, age, mental functioning and task difficulty are relevant factors to consider. In the EFT study by Baron-Cohen and Joliffe (1997) participants were of adult age (mean age 30 yrs; pIQ 105.2) and their mean accuracy scores exceeded the

90% mark, thus falling within the ceiling range, but the authors still recorded faster reaction times in their ASD group. Testing younger participants (mean age 18.8 years; pIQ 98.5), de Jonge, Kemner, and van Engeland (2006) likewise found no differences in accuracy but again recorded faster reaction times for their clinical group, concluding that subtler measures are needed. The earlier study by Ropar and Mitchell (2001) also clearly demonstrated the effects of chronological and mental age, as well as ASD severity on the adult EFT. Their two ASD subgroups consisted of low functioning youngsters with AD (9–18 yrs) and higher functioning children with Asperger Syndrome (8–15 yrs). The AD group outperformed the controls both in accuracy and in reaction times. The Asperger group did not. The mean accuracy score for the controls of the AD group was 25% – far from ceiling.

Nevertheless, our results are still in contrast with those White and Saldaña reported (2011). The most prominent difference between the two studies, is our use of the adult EFT instead of the CEFT in theirs. Moreover, the age ranges of their two ASD samples were wider than the age band of our clinical group (6–12 and 6–16 vs. 9–14 yrs), while their groups also included girls. Yet another important difference might be the variance in task procedures. Unlike in our design, in the White and Saldaña study the target forms were not available during task performance. Consequently, their participants not only had to detect the target forms, they also had to retain them in memory, be able to compare them with other embedded shapes, and distinguish the target forms in the figures, which might then explain their relatively low accuracy scores (61%).

CONCLUSIONS

The results of the present study demonstrate that, if task sensitivity is sufficiently taken into account, boys with ASD do display a subtle superior perceptual processing ability on the Embedded Figures Test compared to typical age peers. In our study both the boys with AD and those with lesser variants of ASD showed a better accuracy on the adult version of the test even though the differences with the controls were small. This superior visual-perceptual capacity is explained by Happé and Frith (2006) as a detail-focused cognitive style, a relevant concept in clinical practice. New research on local processing could shed more light on this style, with the results perhaps guiding practices in the clinical field in new directions.

Acknowledgements

Wim A.J.M. Schlooz was funded by the Reinier van Arkelgroep and the Foundation for Child and Adolescent Psychiatry East Netherlands (Karakter). The authors would like to specially thank Inge Vermeulen for her assistance in testing the children and a first analysis of the data. Without the support of all the children, their parents and schools, this study could not have been done. We thank them for their invaluable help.

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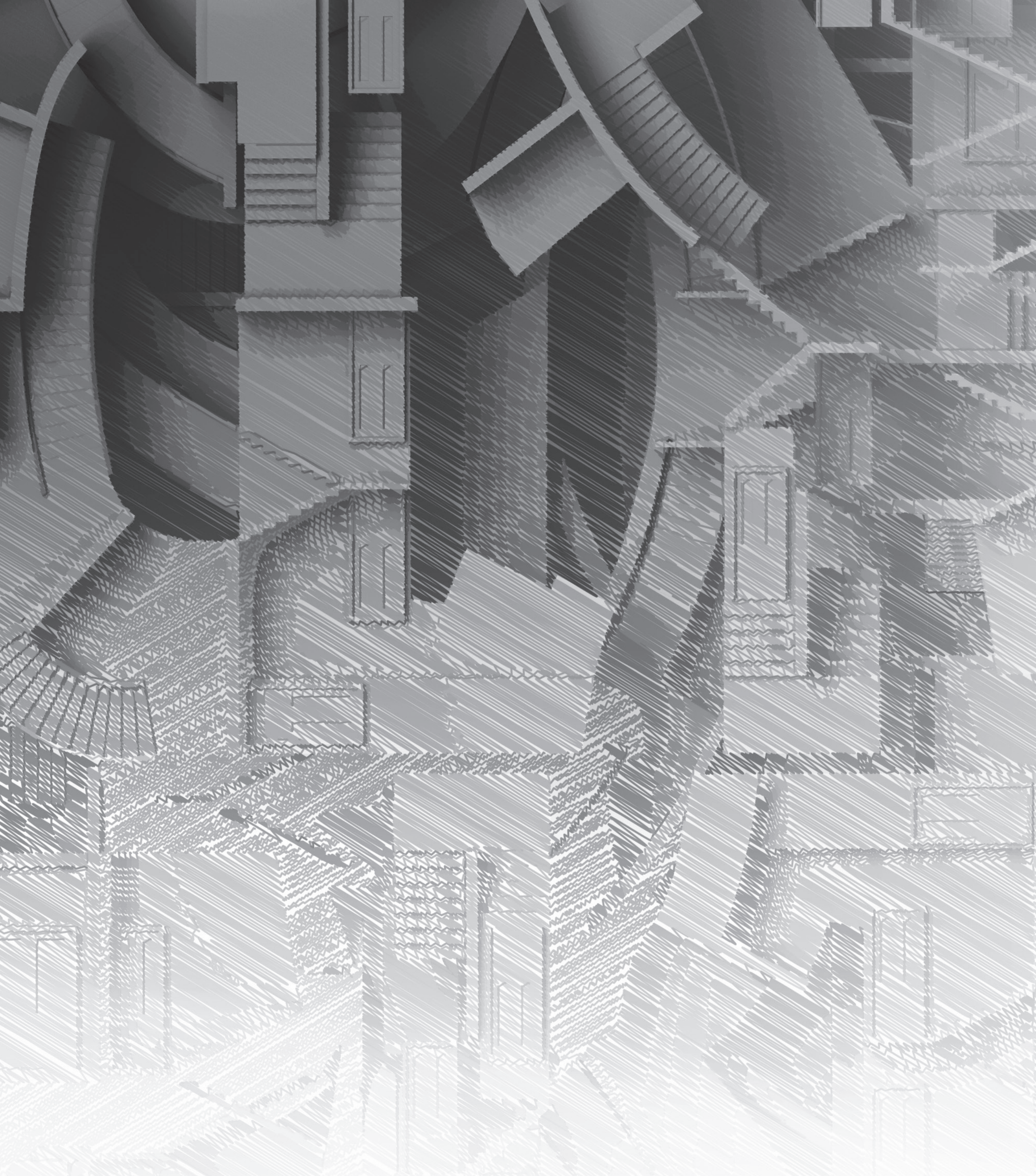

5

Visual completion and complexity of visual shape in children with pervasive developmental disorder

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European Child & Adolescent Psychiatry
(2007), 16, 168-177

ABSTRACT

Much evidence has been gathered for differences in visual perceptual processing in individuals with Autistic Spectrum Disorder. The presence of the fundamental process of visual completion was tested in a group of children with Pervasive Developmental Disorder (PDD), as this requires perceptually integrating visual structure into wholes. In Experiment 1, it was investigated whether visual completion is present for simple partly occluded shapes in a group of children with PDD and a typically developing group. In Experiment 2, the presence of contextual influences in visual completion was investigated for the two groups. A total of 19 children with PDD and 28 controls who were matched for chronological age and IQ took part in two primed-matching tasks. For both groups, visual completion was present and for both groups, contextual influences were found to be dominant in this process. However, only for the group with PDD no priming effects (PEs) were found from less regular primes on congruent test pairs. The group with PDD did integrate visual information into wholes and did this in a contextually dependent way. However, for more complex shapes, visual completion is weaker for this group.

INTRODUCTION

More and more evidence is emerging for differential perceptual processing in individuals with autism compared to typically functioning individuals. Differences in performance in the field of visual perception have been found in various tasks, such as the Embedded Figures Test and the Block Design task (Jolliffe & Baron-Cohen, 1997; Ropar & Mitchell, 2001; Shah & Frith, 1983; Shah & Frith, 1993), the reproduction of impossible figures (Mottron, Belleville, & Menard, 1999), in hierarchical tasks (Plaisted, Swettenham, & Rees, 1999) and in visual search tasks (O'Riordan, Plaisted, Driver, & Baron-Cohen, 2001). Differences also appear in the perception of motion (e.g., on motion coherence tasks (Gepner & Mestre, 2002; Milde et al., 2002) and in biological motion tasks (Blake, Turner, Smoski, Pozdol, & Stone (2003). Although many aspects of vision have been investigated, it is still unclear exactly how visual structure is processed in autism. One typical aspect of autistic' processing is the detail-oriented cognitive style and parallel to this, a deficiency in global processing (Frith & Happé, 1994). This deficiency might extend to a failure to use and integrate visual information, we will therefore look at the ability of a group diagnosed with a disorder in the autistic spectrum to integrate visual information in the so-called process of visual completion. This refers to a basic process in the visual system that allows us to perceive partly occluded objects as whole objects. The presence of this process and contextual influences within this process will be tested using the primed-matching paradigm, a paradigm that has proven to implicitly test visual completion (Sekuler & Palmer, 1992) and is therefore less susceptible to strategies by the participant.

As mentioned, people with autism tend to process information in a detail-oriented way, seemingly at the expense of more global characteristics of the visual world. Most studies have used this notion to look at global and local influences directly, for example by looking at embedded figures, hierarchical stimuli or Gestalt shapes (Brosnan, Scott, Fox, & Pye, 2004; Plaisted, Swettenham, & Rees, 1999; Shah & Frith, 1983; Shah & Frith, 1993). But the findings were not very univocal: initial evidence for a lowered vulnerability to visual illusions, showing a lowered influence of global aspects of an image in autism (Happé, 1996), turned out to be difficult to replicate, the reason of which may be sought in the nature of the instructions (Ropar & Mitchell, 1999). Also, results on hierarchical tasks are mixed. In a hierarchical task introduced by Navon (Navon, 1977), a large letter shape is made up of smaller letters and participants have to identify a letter at the global or at the local level. Usually, people perform better at the global level. For autistic individuals this effect sometimes decreases, but this seems to depend on the type of task that is used, being either a divided or selective attention task (Mottron, Burack, Iarocci, Belleville, & Enns, 2003; Ozonoff, Strayer, McMahon, & Filloux, 1994; Plaisted, Swettenham, & Rees, 1999). Many studies on visual perception in autism have focussed on these global and local aspects, but because higher-level processes, such as strategies, might have influenced performance in some experiments, it is not clear to what level this difference in processing extends.

This study therefore aims to investigate visual structure by looking at a fairly low level of visual integration, namely visual completion, in a group of children with Pervasive Developmental Disorders Not Otherwise Specified (PDD-NOS) and Asperger Syndrome (AS). Autism belongs to the class of Pervasive Developmental Disorders (PDDs) (APA, 1994) and comprises a range of allied disorders, such as autistic disorder (AD), AS, Rett Syndrome, Childhood Disintegrative Disorder, and PDD-NOS. The group of children with PDD-NOS and AS is much larger than the group with AD (Chakrabarti & Fombonne, 2001), and is of great clinical importance. They are treated in clinical practice as if they were suffering from the same anomalies as are found in AD. In addition, there is a high similarity in the epidemiology and etiology in the pattern of AD, AS, and PDD-NOS, and there is evidence for commonalities in visual processing between these three disorders (Schlooz et al., 2006). Since the group of children with (a weaker form of) AD, also have a more detail-based type of processing, this could be reflected in lesser degree of integration (visual completion) or contextual processes could be greatly diminished in visual completion.

Visual completion is a process that we are not aware of: we constantly encounter objects that are partly occluded, but we do not perceive these as fragmented objects, we perceive them as whole objects. For example, the three-quarter of a circle in Figure 5.1A is usually perceived as a circle partly occluded by a rectangle. Alternatively, the left shape in Figure 5.1A can also be perceived as a fragment of a full circle or a mosaic shape, as depicted in Figure 5.1C. As the wholes are the simpler (more regular) shapes, generally, these sorts of configurations are mostly perceived as wholes partly occluded by another shape. This phenomenon is the result of the so-called process of visual completion, which is the ability of the visual system to generate sensations of whole objects from partly occluded objects. The first experimental support for the rapid completion for occluded objects comes from a study on the micro-genesis of visual completion by Sekuler and Palmer (1992). They showed that the percept of partly occluded shapes develops fairly fast into the percept of a whole shape, taking about 250 ms to develop. Using a visual search task, Rensink and Enns (1998) showed that partly occluded shapes are harder to find among complete shapes as compared to parts, indicating that partly occluded shapes are perceived as being more similar to complete shapes, again showing the fairly low

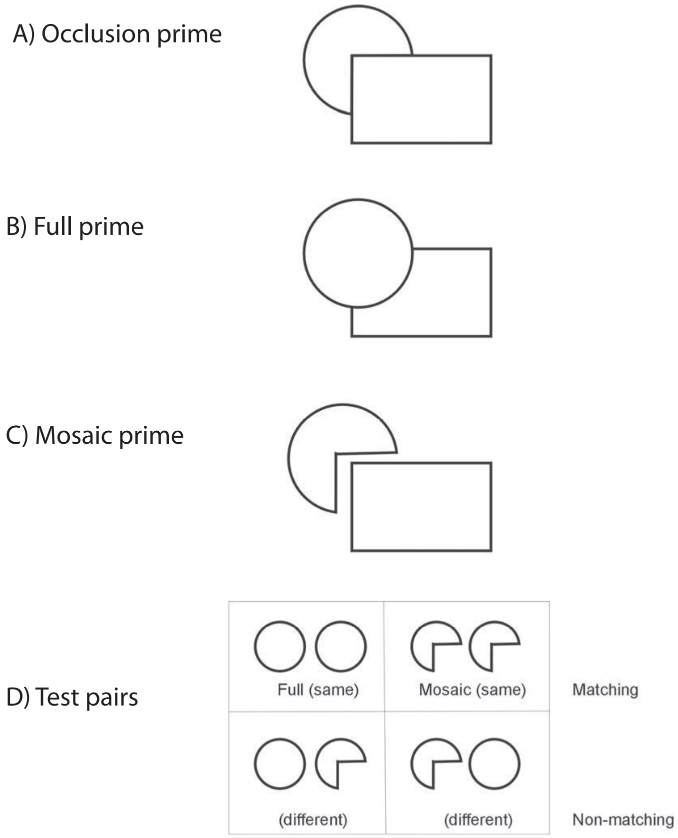


Figure 5.1 A, B and C are primes. **(A)** The shape on the left is partly occluded by the rectangle, the occlusion prime. **(B)** Completion of the partly occluded shape as predicted by most theories, the full prime. **(C)** A mosaic version of the partly occluded shape, the mosaic prime. **(D)** The test pairs.

level at which visual completion operates. Furthermore, in an fMRI study by Kourtzi and Kanwisher (2001) a pair of identical shapes was shown sequentially where one shape was in front of a grid of parallel bars, and the other was occluded by the same bars. Therefore, the contours were different, but participants perceived the same shape. Compared to a condition in which this was reversed (same contours, but different perceived shape) they found a smaller response in the lateral occipital complex (LOC). This indicates that perceived shapes and not contours per se, are represented in a visual area such as the LOC. In addition, visual completion is a process that is already present in 4-month-old infants (Kellman & Spelke, 1983).

Besides the perception of a whole shape, we even have a strong sense of the form of the occluded part. This perceived form can either be based on overall contextual information,

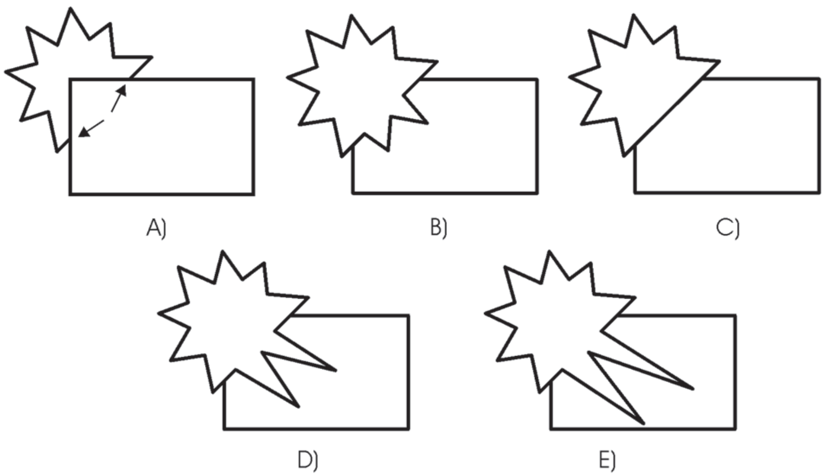


Figure 5.2 **(A)** The shape on the left from set 1 is partly occluded by the right shape. **(B)** Contextual (global) completion, resulting in a simple regular overall shape. **(C)** Local completion, resulting in a less regular (more complex) overall shape. **(D)** Set 2. **(E)** Set 3.

or can just be based on information that is available at the intersections between the partly occluded shape and the occluder (see the arrows in Figure 5.2A). Contextual information originates from the whole visible part of the partly occluded shape, and include stimulus aspects such as symmetry and iteration, see Figure 5.2B (Van Lier, 1999; Van Lier, Van der Helm, & Leeuwenberg, 1994; Van Lier, Van der Helm, & Leeuwenberg, 1995). Completions in which contextual information plays a role, can take any kind of form, depending on the structural properties of the visible part. By definition this results in the most regular or simple shape. Completions that are just triggered by intersections always proceed by way of linear or smooth continuation of the occluded contours, see Figure 5.2C for an example (Fantoni & Gerbino, 2003; Kellman & Shipley, 1991). It is now generally agreed upon that the perceived completion depends on the nature of the stimuli such as the type of regularities or the degree of occlusion (Sekuler, 1994; Van Lier, Van der Helm, & Leeuwenberg, 1995; Van Lier & Wagemans, 1999). For example, for the kinds of shape shown in Figure 5.2A, earlier experiments with adult participants showed that contextual influences dominate visual completion (De Wit & Van Lier, 2002; De Wit, Bauer, Oostenveld, Fries, & Van Lier, in press; De Wit, Mol, & Van Lier, 2005). Unfortunately, to date there is no direct neurophysiologic substrate for visual completion nor for these intersectional and contextual influences. However, in an MEG study it was shown that there is an early left occipital component that is mainly sensitive to pure figural aspects, but that is already modulated by these global influences (De Wit et al., in press), this is line with an EEG study showing that the recognition of partially visible objects takes place relatively early in the visual stream (Johnson & Olshausen, 2005).

Before investigating contextual influences in visual completion in PDD, we wanted to see if a difference exists in visual completion for simple shapes (in which both intersectional and contextual influences result in the same completions) between children and adults. Following earlier research on visual completion, we used the primed-matching paradigm, which can be used to implicitly test how different images are perceived. The paradigm has been extensively used to explore visual completion in adults (De Wit & Van Lier, 2002; De Wit, De Weert, & Van Lier, 2005; De Wit, Mol, & Van Lier, 2005; Sekuler, 1994; Sekuler & Palmer, 1992), but has not yet been tested in children. In this paradigm, the participant's task is to decide whether a test pair consists of two identical shapes, or not. A test pair is termed to be "matching" if both shapes in the test pairs are identical and "non-matching" if this is not the case. In case of a matching test pair, the decision on similarity can be facilitated by showing a similar prime before this test pair (Beller, 1971). The rationale behind the paradigm is that the facilitation depends on the perceived similarity between the prime and the two shapes in the test pair. When investigating visual completion, a partly occluded shape is shown as a prime ("occlusion prime"), which will speed up the reaction to a test pair that is perceived as being similar. The pattern in Figure 5.1A can be seen either as a mosaic or a complete circle partly occluded by the rectangle. So the effect of the occlusion prime can be tested on a test pair containing either two mosaic forms of the shape ("mosaic test pair") or by two completed forms of the shapes ("full test pair"), see Figure 5.1. When the occlusion prime facilitates a mosaic test pair most, the partly occluded shape is perceived as a mosaic shape. When full shapes are facilitated most, the partly occluded shape is perceived as complete shape, reflecting that a completion process did take place.

The performance of the group with PDD will be compared to a normally developing group that is matched on age and intelligence. For both groups we expect general priming effects (PEs) from full primes on full test pairs, and mosaic primes on mosaic test pairs. For the occlusion prime, we expect a PE on the full test pairs in the control group. If visual completion is weaker in the group with PDD, we expect a smaller PE on the full test pair, as compared to the control group, or even a PE on the mosaic test pair. After testing the paradigm for this age group and comparing performance on simple shapes for the PDD group and the control group, the importance of contextual influences will be investigated in Experiment 2.

EXPERIMENT 1

METHOD

Participants

A total of 19 children with PDD-NOS (16) and AS (3) were recruited from regional outpatient clinics for Child Psychiatry (GGZ Nijmegen) associated with the outpatient Clinic of Child Behavioral Neurology and the Academic Centre for Child and Adolescent Psychiatry (ACKJON) of the University Medical Centre St. Radboud, Nijmegen, the Netherlands. Participants had been diagnosed by experienced clinicians, child psychiatrists and child psychologists both independently and together. The most effective scoring rule for PDD-NOS based on ICD 10/DSM-IV criteria (APA, 1994; Buitelaar & Van der Gaag, 1998; Buitelaar, Van der Gaag, Klin, & Volkmar, 1999) was applied, i.e., a short set of seven criteria that have all been derived from the original twelve criteria for AD defined in the DSM-IV. The threshold for inclusion in the PDD group was set at three out of seven criteria, of which at least one needed to be in the social interaction domain (also reported in Schlooz et al., 2006). The group did not contain any children with autism Children with co-morbidity were excluded. At the time of testing, none of the participants were using medication. A total of 28 healthy primary schoolchildren were selected from two primary schools to form the age- and intelligence matched control group. All participants were male, had normal or corrected-to-normal vision and functioned on an average cognitive level. The mental functioning of all participants was assessed with the Wechsler Intelligence Scale for Children—Revised, WISC-R (Wechsler, 1974). Table 5.1 presents the chronological age and performance on the WISCR for the two groups.

Table 5.1 Chronological age and performance on the Wechsler Intelligence Scale for Children—Revised, between brackets standard deviation

Group	Age (years:months)	TIQ	VIQ	PIQ
Control group	11:0 (0.7)	108.1 (10.5)	106.9 (11.3)	107.1 (10.7)
PDD group	12:2 (1.7)	106.1 (14.1)	105.3 (15.9)	106.1 (14.4)

Stimuli

Three sets of stimuli were used, consisting of basic shapes (circles, squares, and rectangles). To check the presence of a general PE, there also is a control condition in which mosaic shapes and the full shapes serve as primes (as these are identical to the mosaic test pair and the full test pair respectively, PEs should appear in these conditions). These conditions will also be used to compare with the effect of the occlusion prime. Therefore there were three possible configurations: (1) the shapes could either be occluded by a rectangle (the occlusion primes), or (2) the full shapes shown in front of the rectangle (the full primes), or (3) the shapes formed a mosaic version of the occlusion prime, where the shapes were displaced with respect to the rectangle (see Figure 5.1 for the primes and test pairs). The visual angle of the prime was 1.75°. A dot functioned as the no-prime condition.

The test pair consisted of two shapes of the same set as the prime. The task for the participants was to judge if the two shapes in the test pair were the "same" (matching) or "different" (non-matching) with respect to each other. There was either a matching test pair with two completed, full shapes, or two mosaic shapes, or a non-matching test pair. A non-matching test pair was a combination of a completed shape and a mosaic shape from the same set, and was controlled for left–right position. The matching test pairs were shown as often as the non-matching test pairs. The shapes in the test pair appeared on both sides of the prime. Note that the positions of the shapes were located away from the prime to inhibit masking by the prime, see Sekuler and Palmer (1992). The right shape of the prime appeared on the lower part of the screen. This inhibited the illusion of the rectangle moving and changing into one shape of the test pair, which could also exercise a hindering influence on the PE, also see Sekuler and Palmer (1992). The experiment was run with Presentation, version 5.2 (Neurobehavioral Systems).

Procedure

The clinical group was tested in the hospital. The control group was tested in their school. The same setup was used for both groups (same computer screen and same viewing distance). A trial started with a fixation cross being presented in the middle of the screen for 500 ms. After a blank screen was shown for 50 ms, a prime appeared on the screen for 500 ms. After a 17 ms interstimulus interval, the test display was shown until the participant responded with a button press (Figure 5.3). To respond to a matching test pair, the right index finger was used to press the right button, to respond to a non-matching test pair, the left index

Figure 5.3 Procedure of the primed-matching paradigm as used in Experiments 1 and 2.

finger was used to press the left button. The order of the presentation was randomized for each participant and the reaction time (RT) was measured to the nearest millisecond. The trials were presented in eight blocks of 36 continuous trials, and after every block a pause was given. After each block, feedback was provided on the number of correct responses during this block.

To minimise attentional effects participants were instructed to pay extra attention to the left shape of the prime (because variations of this shape followed in the test pair) and to respond as accurately and as fast as possible when the test pair appeared on the screen. The experiment started with eight practice trials in which feedback was also provided. The experimental trials consisted of: set (3) · primes (4: occlusion, full, mosaic, and no-prime) · test pair (4: full (matching), mosaic (matching), full-mosaic (non-matching), and mosaic-full (non-matching)) · repetition (4). Note that, occlusion prime and no-prime conditions were shown twice as often, resulting in 288 trials.

RESULTS

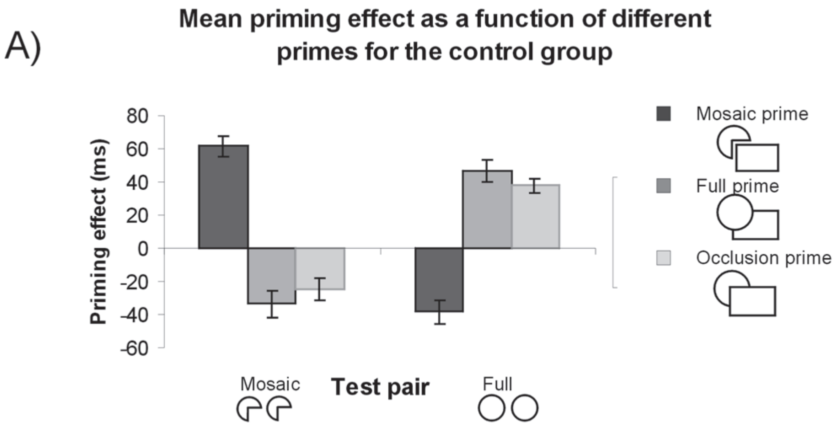
All correctly answered matching test pairs were analyzed (95.94% for the control group and 95.82% for the PDD group; the pattern of errors was similar between groups), see Table 5.2 for mean RTs and standard error of the mean (SEM). This has been done because PEs only occur for "same" test pairs, as "different" test pairs do not show a regular pattern (Beller, 1971; Sekuler & Palmer, 1992). Responses shorter or longer than 2SD from the mean for each participant's responses (by participant) were removed from the analyses. First, note that the RTs for the full test pairs and the mosaic test pairs in the no-prime conditions did not differ between the two groups ($t(45) = 0.640$, $P = 0.525$ and $t(45) = 0.112$, $P = 0.911$, respectively). Therefore, differences between groups are specific for the PEs.

The PE is defined as the difference in RT between a prime condition and a no-prime condition: PE (Test Pair | Any Prime) = RT (Test Pair | No Prime) – RT (Test Pair | Any Prime).

Table 5.2 Mean RTs (ms) and standard errors of the mean between brackets for all conditions and matching test pairs in Experiments 1 and 2

Prime	Control group		PDD group	
	Full test pair	Mosaic test pair	Full test pair	Mosaic test pair
Experiment 1				
Occlusion	628.52 (34.52)	785.92 (42.42)	664.33 (41.64)	800.46 (50.98)
Full	619.62 (33.17)	795.06 (44.85)	658.03 (39.98)	801.70 (53.85)
Mosaic	704.36 (34.90)	699.68 (37.67)	742.47 (42.00)	739.67 (44.03)
No-prime	666.18 (37.13)	761.27 (36.64)	703.56 (44.65)	754.81 (43.99)
	Local test pair	Global test pair	Local test pair	Global test pair
Experiment 2				
Occlusion	862.09 (36.29)	719.03 (30.69)	881.90 (44.05)	782.05 (37.26)
Local	795.27 (36.72)	862.61 (36.85)	821.06 (44.57)	845.73 (44.74)
Global	899.83 (38.75)	705.76 (32.61)	935.41 (47.05)	748.00 (39.59)
No-prime	868.29 (35.77)	776.00 (34.06)	852.58 (43.42)	820.20 (41.35)

In Figure 5.4A, mean PEs are plotted for the control group averaged over all test pairs as a function of type of prime, in Figure 5.4B this is plotted for the PDD group. A repeated-measures ANOVA was performed for PE with Prime (3: occlusion, full, and matching), and Test Pair (2: full and mosaic) as within subjects variables and Group (2: PDD and control) as a between subjects variable. This revealed a significant main effect for Test pair $F_{1,45} = 4.633$, $P < 0.05$, where full test pairs were primed more than mosaic test pairs (also RTs for full test pairs were lower than for mosaic test pairs, $F_{1,45} = 140.240$, $P < 0.001$). Furthermore, an interaction effect was found for Prime · Test pair, $F_{2,44} = 36.818$, $P < 0.001$. Planned comparisons were performed to investigate the PE of occlusion primes on full test pairs compared with the no-prime condition; this was significant for both groups (Control: $F_{1,27} = 15.301$, $P < 0.005$; PDD: $F_{1,18} = 13.402$, $P < 0.001$). Also, the effect of full primes on full test pairs was significant for both groups (Control: $F_{1,27} = 13.182$, $P < 0.005$; PDD: $F_{1,18} = 6.308$, $P < 0.05$). However, the effect of the mosaic prime on the mosaic test pairs was only significant for the control group ($F_{1,27} = 20.138$, $P < 0.001$), not for the PDD group ($F_{1,18} = 1.429$, $P = 0.247$). To test for group differences, a planned comparison was performed for an interaction between the effect of the mosaic prime and group and this was significant, $F_{1,45} = 5.572$, $P < 0.05$.



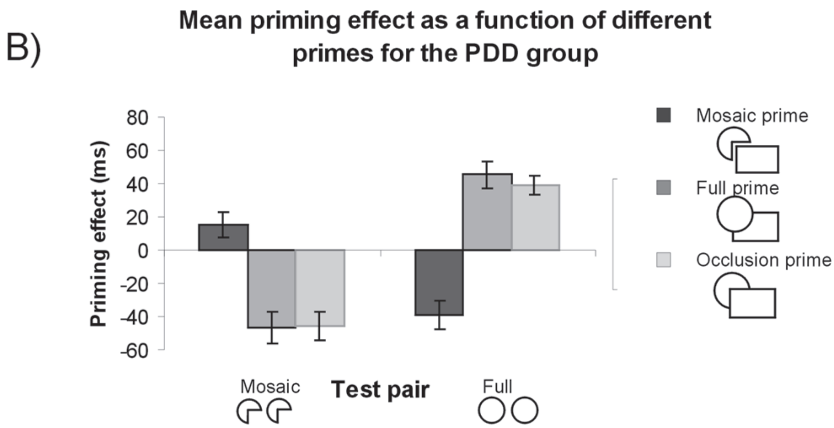


Figure 5.4 Mean priming effect for the test pairs as a function of all primes for the control group (panel **A**) and for the PDD group (Panel **B**). The bars represent mean standard error.

DISCUSSION

Both groups do not differ from each other in the speed of reactions, which indicates that possible differences between the two groups are not caused by a difference in overall speed. For the control group, a PE was found for full primes on full test pairs and for mosaic primes on mosaic test pairs, which fits in with earlier findings on adults (Sekuler & Palmer, 1992). Thus, the basic PEs are present, showing that this paradigm is a valid method of investigating visual representations in this age group. As in earlier data from adults, these data also show that the occlusion primes facilitate performance on full test pairs, but not on mosaic test pairs, reflecting the presence of the visual completion in the control group. To our knowledge, the present study is the first to show that this sort of priming is already present for this age group. For the PDD group we found a similar pattern: occlusion primes and full primes also facilitate complete test pairs. This signals an intact visual completion process for the PDD group for these simple shapes, so the PDD group integrates visual information equally well into wholes. However, there is a difference between the PDD and the control group: the PE of the mosaic prime on the mosaic test pair is nearly absent in the PDD group. One way to explain this is to think of the complete circle as a simple (regular) shape. The mosaic could be interpreted as being a deviation of a simple (regular) circular shape. This might indicate that the PDD group has a difficulty in dealing with more complex shapes. We will return to this issue later on.

As it is clear that the PDD group can integrate information into wholes, we will turn to the question of the perceived form of occluded parts and see if contextual influences also play a role in visual completion in PDD.

EXPERIMENT 2

To investigate the effect of context in visual completion, stimuli from earlier experiments on visual completion were used (De Wit & Van Lier, 2002; De Wit, Mol, & Van Lier, 2005). As in Experiment 1, the effect of an occluded shape was tested, but the shapes used in this experiment can give rise to qualitatively different completions. Take for example the shape in Figure 5.2A, which can be completed in two plausible ways. The completion can either be triggered by contextual information resulting in a regular (simple) shape, which we will refer to as a *global* completion (Sekuler, 1994; Van Lier, Van der Helm, & Leeuwenberg, 1994; Van Lier, Van der Helm, & Leeuwenberg, 1995) (see Figure 5.2B), or the completion can solely be triggered by information at the location where the partly occluded shape meets the occluder, resulting in a linear or curved continuation of the partly occluded contours, which we will refer to as a *local* completion (Kellman & Shipley, 1991) (see Figure 5.2C). For the shapes used here, this latter completion is more complex in terms of regularity. Earlier experiments with the shapes used in this experiment showed that contextual influences dominate for adults (De Wit & Van Lier, 2002; De Wit et al., in press; De Wit, Mol, & Van Lier, 2005). That is, for the current shapes, global completions were preferred compared to local completions. As addressed in the introduction, there is a tendency in autistic individuals to be more focussed on details, seemingly at the expense of more global characteristics of the visual world. In line with this, we expect the PDD group to be less sensitive to contextual influences. For the control group we expect visual completions to be similar to that of adults, and therefore be dominated by contextual influences.

METHOD

Participants

The groups of participants were identical to Experiment 1.

Stimuli

Three sets of stimuli were used (Figure 5.2A, D, and E), already used by De Wit et al. (2002). The shapes could either be occluded by a rectangle (the occlusion primes) or full shapes in front of a rectangle. The full primes comprised locally or globally completed shapes (i.e., the local and global prime, respectively) that were positioned in front of the rectangle. In the local prime, the contours that disappear behind the occluder in the occluded version are linearly continued until the lines meet. In the global prime, the same kind of protrusions available in the visible part were continued in the completion. The visual angle of the prime was 1.52°. A dot functioned as the no-prime condition.

Prime and test pair were drawn from the same set. This was either a matching ("same") test pair with two global or two local shapes or a non-matching ("different") test pair. Matching test pairs were shown as often as the non-matching test pairs. The test pairs appeared on both sides of the prime, to inhibit masking by the prime, as in Experiment 1. Also, the right shape of the prime appeared on the lower part of the screen.

Procedure

The procedure was identical to Experiment 1, again there was a total of 288 experimental trials: set (3) · primes (4: occlusion, local, global, no-prime) · test pair (4: local–local, global–global, local–global and global–local) · repetition (4). Note that occlusion prime and no-prime conditions were shown twice as often.

RESULTS

All correctly answered matching test pairs (95.14% for the control group and 96.0% for the PDD group) were analyzed, see Table 5.2 for mean RTs and SEM. Responses shorter or longer than 2SD from the mean for each participant's responses (by participant) were removed from the analyses. Note again that the RTs for the local and global test pairs in the no-prime conditions did not differ between the two groups ($t(45) = 0.528$, $P = 0.781$ and $t(45) = 0.908$, $P = 0.414$, respectively). Therefore, differences between groups are specific for the PEs. In Figure 5.5A, mean PEs are plotted for the control group for all test pairs as a function of all primes, in Figure 5.5B these are plotted for the PDD group. A repeated-measures ANOVA was performed for PE with Prime (3: occlusion, global, and local), and Test Pair (2: global and local) as within subjects variables and Group (2: PDD and control) as a between subjects variable. This revealed a marginally significant main effect for Test pair ($F_{1,45} = 3.773$, $P = 0.058$), and a significant interaction between Test pair · Group ($F_{1,45} = 4.477$, $P < 0.05$). Also, a significant interaction effect was found for Prime · Test pair ($F_{2,44} = 57.332$, $P < 0.001$). Furthermore, a three-way interaction effect was found for Prime · Test pair · Group, $F_{2,44} = 3.666$, $P < 0.05$.

Planned comparisons were performed to investigate the PE of occlusion primes on local and global test pairs compared with the no-prime condition. The occlusion prime had no effect on local test pairs for both groups (Control: $F_{1,27} = 0.529$, $P = 0.473$; PDD: $F_{1,18} = 2.831$, $P = 0.110$), but it did have a significant PE on global test pairs for both groups (Control: $F_{1,27} = 26.628$, $P < 0.001$; PDD: $F_{1,18} = 7.324$, $P < 0.05$). Also, the effect of global primes on global test pairs was significant for both groups (Control $F_{1,27} = 26.314$, $P < 0.001$; PDD: $F_{1,18} = 23.486$, $P < 0.001$). The effect of the local primes on the local test pairs was significant for the control group ($F_{1,27} = 33.554$, $P < 0.001$) but not for the PDD group ($F_{1,18} = 2.094$, $P = 0.165$). To test for group differences, a planned comparison was performed for an interaction between the effect of the local prime and test pair and group but this was marginally significant, $F_{1,45} = 3.107$, $P = 0.085$. There was a difference between the groups in the magnitude of the PE of the occlusion prime on the global test pair. In the control group, the size of this PE did not differ from the PE of the global prime on the global test pair ($t(27) = 1.060$, $P = 0.298$), whereas in the PDD group this PE of the global prime on the global test pair was significantly larger than the PE of the occlusion prime on the global test pair ($t(18) = 3.085$, $P < 0.01$). To test for group differences, a planned comparison was performed for an interaction between the effect of the occlusion prime and global prime on global test pair between groups and this was not significant, $F_{1,45} = 1.375$, $P = 0.247$.

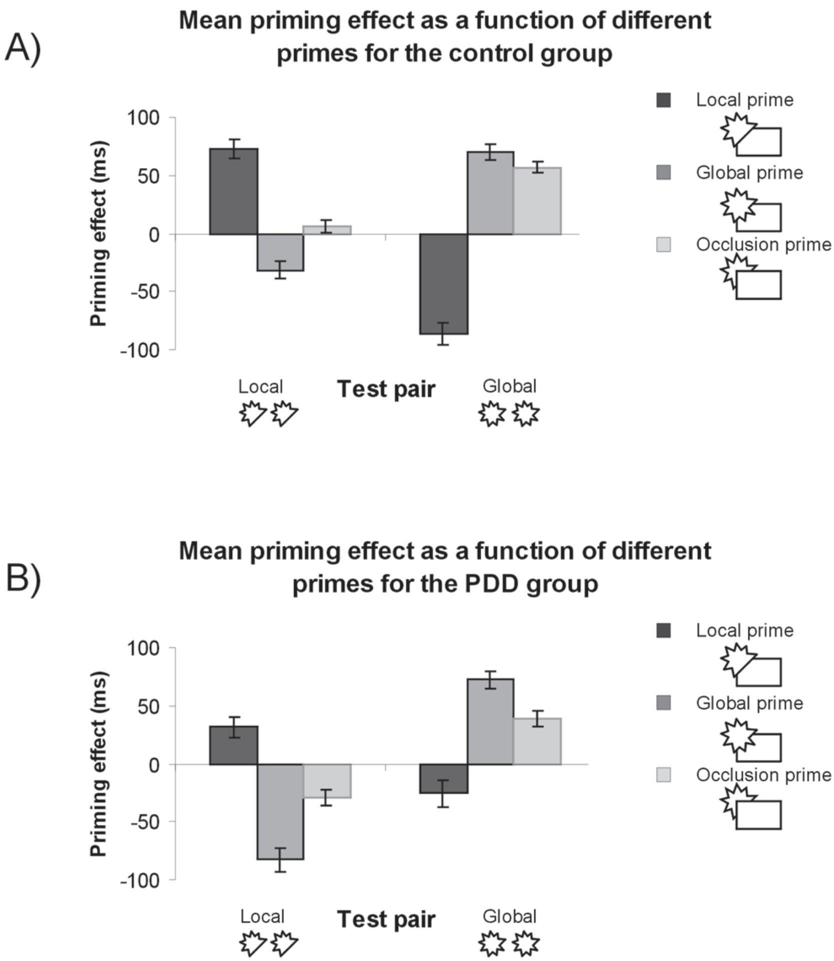


Figure 5.5 Mean priming effect for the test pairs as a function of all primes for the control group (panel **A**) and for the PDD group (panel **B**). The bars represent mean standard error.

DISCUSSION

For the control group it was found that local primes facilitate local test pairs and global primes facilitate global test pairs. Moreover, the occlusion primes facilitate the global test pairs, but not the local test pairs, similar to what has been found for adults (De Wit & Van Lier, 2002; De Wit et al., in press; De Wit, Mol, & Van Lier, 2005). For the PDD group a similar pattern was found for the occlusion and global primes: both facilitate global test pairs. Therefore, contextual influences in visual completions are apparent for both groups. However, there are three notable differences between the two groups.

First, the strength of the PE of occlusion primes on global test pairs differs between the control and the PDD group. For the control group the PE of the occlusion primes on the global test pairs is the same as the effect the global primes have on these global test pairs. However, for the PDD group the effect of the occlusion primes is significantly smaller than the effect of the global primes. Although this is not reflected in the test for group differences, these data suggest that visual completion for these stimuli is not as robust for the PDD group as it is for the control group.

Second, in the control group local primes do facilitate local test pairs, whereas in the PDD group, local primes do not facilitate local test pairs (the group comparison was marginally significant, $P = 0.085$). Note that there is facilitation from the regular (global) primes on regular test pairs in the PDD group. Although the absence of facilitation for the less regular (local) shapes does not relate to the process of visual completion directly, it does suggest a difference in the processing of visual structure which is modulated by the complexity of the shapes. This is analogous to the finding in Experiment 1, where mosaic shapes, being more complex than the completed (full) circles, also lack a facilitating effect.

Third, comparing the two groups in Figure 5.5 reveals another difference between controls and individuals with PDD. In the controls, local primes exert a large inhibiting effect on global test pairs, while global primes do not have such an inhibiting effect on local test pairs. This effect is reversed in the PDD group, here the local primes do not effect global test pairs, but the global primes inhibit reaction to the local test pairs ($F_{1,45} = 7.191$, $P < 0.05$). To explain these clear differential effects between the two groups we can only speculate. For example, when considering the irregular (local) shape to be a deviation of the regular (global) shape, the pattern of results suggests an asymmetry with regard to pattern classification. For the PDD group a deviation from a simple, regular shape seems to be quite unrelated, whereas for the control group this is the other way around. This asymmetry might point to a difference in dealing with regular shapes and deviations of these shapes. It should be noted, however, that that the above suggestion calls for further investigation (e.g., dealing with recognition of prototypical structures versus subordinate structures). As this finding does not deal with visual completion per se we consider a full account beyond the scope of this article. Nevertheless, the results again show a significant difference between the PDD group and the control group obtained with the primed-matching paradigm.

GENERAL DISCUSSION

The presence of PEs in both experiments shows that the primed-matching paradigm is a suitable method to investigate visual perception in children in an implicit way. Mean RTs are not different for the two groups, therefore the results cannot simply be explained by a general slowing-down or speeding-up, but results are specific to the PE itself, indicating a different way of visual processing. Initially, these data suggest that the process of visual completion in PDD does not differ from visual processing in the control group. The PDD group also perceives partly occluded shapes as whole shapes, and the fact that the experienced form of the completions is the same as for the control group shows that this completion process can also be influenced by contextual factors. Inspecting the data more closely however, reveals two main effects that do differentiate between individuals with PDD and controls.

One is a difference specific to visual completion and the second is the lack of PE in certain conditions. The first indication for a difference between the two groups comes from the data of Experiment 2, which show that the visual completion process in the PDD group is present, but it is weaker as compared to an equivalent full (completed) prime. This finding differs from the results of Experiment 1, where we did not find a difference between the PE of partly occluded primes versus full primes between the groups when comparing completed shapes and mosaic shapes. The difference in results between these two experiments may lie in the fact that both intersectional and contextual information trigger the same (circular) completion in Experiment 1. However, in Experiment 2 they trigger different completions, resulting in a more ambiguous condition. In addition, in the difference in overall complexity of the shapes that were used in Experiment 2 as compared to the shapes in Experiment 1. That is, more complex stimuli reveal subtler group differences in completion processes in PDD.

The second indication for a difference between the two groups lies in the effect of the less regular shapes found for the PDD group in both experiments. Specific shapes (the mosaic shapes in Experiment 1 and the “local” shapes in Experiment 2) did not facilitate identical test pairs. This could mean that it is harder for the PDD group to process these irregular, more complex stimuli (as regularity is an important factor in object representation (De Wit, Mol, & Van Lier, 2005; Van Lier & Wagemans, 1999)). This might relate to the suggestion by Plaisted et al. (1998) on enhanced discrimination in autism. In a study on perceptual learning they showed that high-functioning autistic individuals were equal to controls at discriminating familiar objects, but better than controls at discriminating novel objects, suggesting an absence of a perceptual learning effect. In the most regular shapes, which are also more familiar, in our experiments there were no differences between the groups. Differences arose when irregularity came in, there the PDD group might have more difficulty in processing those shapes in the same way as regular (more familiar?) shapes.

It is remarkable that a group of children with a lesser variant of PDD also appears to differ from typical developing children in the way they process visual stimuli even at a fairly low level of perception. This is in line with findings by Schlooz et al. (2006) that showed fragmented visuo-spatial processing in this group, although not much more is known on performance of this group, it does imply a commonality between different PDD groups.

How do our findings relate to theories on perception in autism? Our findings do not directly confirm the Weak Central Coherence theory (Frith, 1989), as we did find contextual influences in completions. However, WCC was refined by Happé (1999) by suggesting that individuals with autism are not impaired in the ability to integrate the various features and facets that make up a single object. Although this does agree with our data on simple partly occluded shapes, we do find a problem in processing more complex partly occluded stimuli. These more complex stimuli are likely to reflect less known categories in the visual world. In that sense, visual processing in PDD can be regarded as being more inflexible in dealing with new information. Another model of visual processing in autism is the Enhanced Perceptual Functioning model by Mottron and Burack (2001), which states that the overdevelopment of low-level perceptual operations might result in an atypical relationship between global and local processes. This model is similar to the notion of enhanced discrimination by Plaisted et al. (1998) and our findings also fit in with this notion. An alternative idea on why visual processing in autism is different suggests that autistic individuals are less influenced by prior knowledge (Ropar & Mitchell, 2002). Our results show that prior knowledge (or temporal context) in the form of the prime does influence performance, although we also find that it depends on the sort of shape (e.g., for the mosaic shapes this effect seems diminished). The difference in processing of visual structure in autism seems not to be in binding features into whole shapes, but rather at a more complex (categorical) level of perception.

CONCLUSIONS

Visual completion was apparent in the PDD group we tested and similar to the control group, contextual influences are apparent in the completion process for the stimuli that were used. However, complexity of the shape exerted an effect on the strength of visual completion and there also was a difference between the groups in the effect of regularity in shapes: the PDD group shows reduced effects from less regular shapes, also reflecting an effect of complexity.

Acknowledgements

TdW was funded by the Netherlands Organisation of Scientific Research (NWO) on a project awarded to RvL, WS by the Foundation for Child and Adolescent Psychiatry East Netherlands (SKJPON, Stichting Kinder- en Jeugdpsychiatrie Oost Nederland) and RvL received a grant from the Royal Netherlands Academy of Arts and Sciences. The authors would like to thank two anonymous reviewers for their helpful comments, Ilse Groenendaal en Anne Monsuwe for their assistance in testing the children and of course all children, their parents and schools for their invaluable help.

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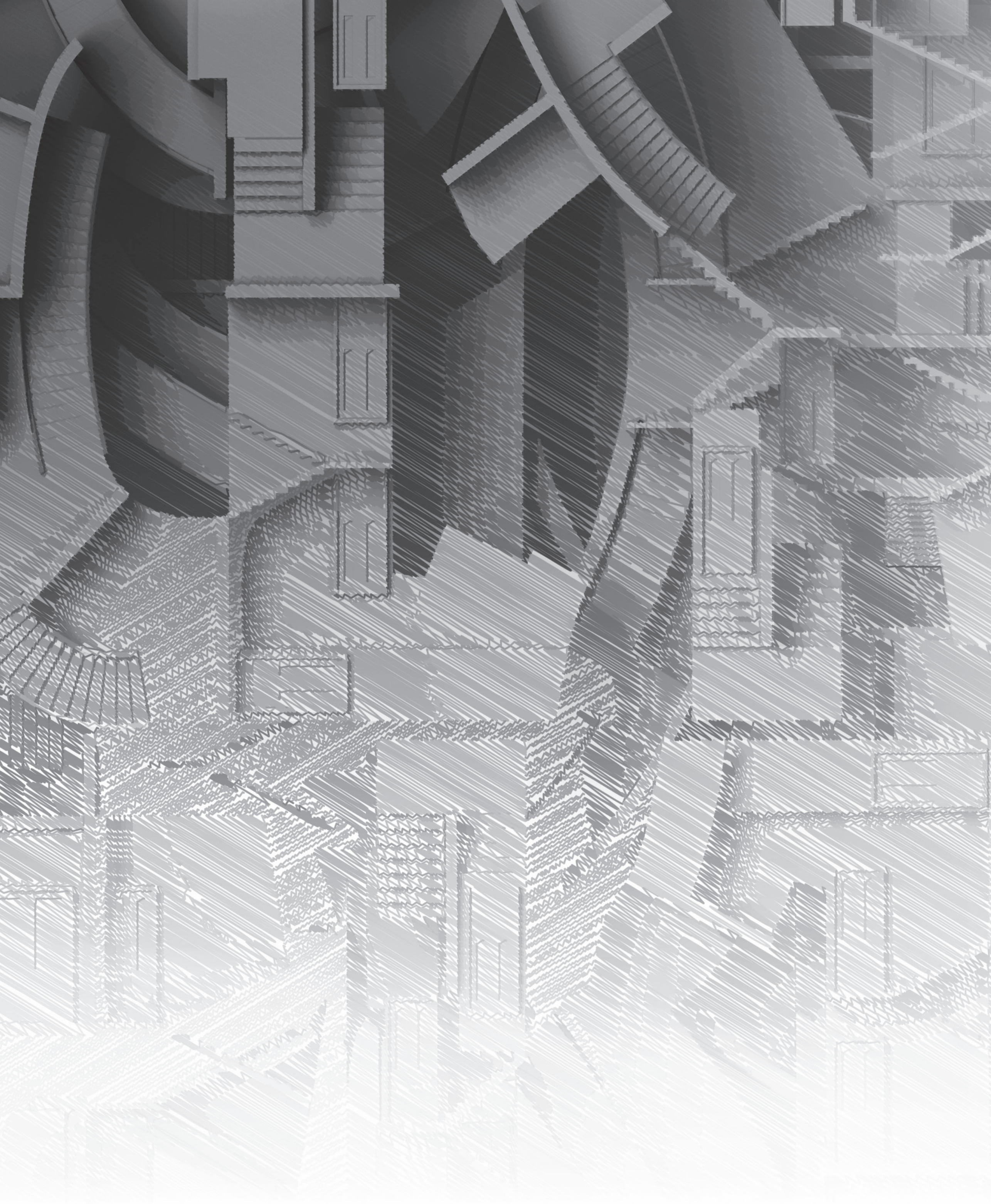

6

Extended summary and discussion

Children living with autism spectrum disorder (ASD) have serious problems communicating and interacting socially and show restricted, repetitive patterns and behaviours (DSM 5, APA 2013). Their deficits are severe and pervasive in nature. Many domains of functioning are affected: initiating and maintaining social contacts, speech and language, planning and organisation, and often also motor skills. In all stages of their lives, they will be facing problems and challenges in these areas. Their neurocognitive mode of functioning is quite distinctive and of great clinical relevance. Systematic observations of the way these youngsters behave and function have offered us insight into their weaknesses and strengths, have helped us better understand what burdens and impairs them and which conditions help them thrive.

The Diagnostic and Statistical Manual of Mental Disorders (DSM) is an important classification benchmark for psychiatrists, comparable with the International Statistical Classification of Diseases and Related Health Problems (ICD, World Health Organization, 1993). At the time of writing this thesis, both the DSM-IV (APA, 1994) and the DSM-5 (APA, 2013) classification systems prevailed. In this thesis the reader has hence come across designations derived from both classifications, where the children with the severest ASD type were denoted as suffering from autistic disorder (AD) and those diagnosed with milder variants of ASD as having Asperger syndrome or Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS), with the latter two being labelled as the 'PDD' group.

Although motor ability is not part of the criteria for an ASD diagnosis, the children's motor development does show typical tendencies (Fournier, Hass, Nail, Lodha, & Caurugh, 2010). Apart from being delayed in their development, they often take (much) longer to learn to ride a bike or swim and at elementary school tend to lag (seriously) behind in learning to write (Kushki, Chau, & Anagnostou, 2011).

Clinical observation suggests that children with ASD have problems in drawing a person as compared to typically developing children (Chapter 1). Analysing sketches of a human person drawn by young children and quantifying their problems in drawing is, however, very complex. In addition, these difficulties in drawing might not be a pure motor problem but could also result from deficits in their ability to build and apply higher order concepts such as body representation (Cascio, Foss-Feig, Burnette, Heacock, & Cosby, 2012) in motor control. Nevertheless, drawing tasks are a very good means to study the role and effects of higher order visual perceptual concepts on motor performance. For methodological reasons, in our research we refrained from analysing free drawings (for example sketches of a person) and exploited familiar and validated visuomotor tests, most specifically drawing tasks. By doing so, we expected to gain a better understanding of the visuomotor functioning of children diagnosed with a milder ASD and to establish whether they share the same deficiencies observed in peers with severe ASD as compared to their typically developing counterparts.

Three guiding neurocognitive theories, the social cognition, the defective executive function, and the central coherence accounts, each propose specific deficits to underlie autism. These three models have been discussed in Chapter 1, where it was argued why in this dissertation the main focus was on the latter account. The central coherence hypothesis (Frith, 1989; Happé & Frith, 1996) proposes that people with autism have a strong detail-focused tendency and make little use of context or existing structures in which component parts are embedded: they are masters in detail, with little eye for overall structures. Francesca Happé (1999) speaks of "a cognitive style biased towards local rather than global information processing". This cognitive-functional mode clearly has its drawbacks. In order to be able to get the overall picture, this global picture is not directly 'seen' but has to be constructed by exploring all its constituent components (Kanner, 1943). This not only takes time, but also renders children and adults with ASD vulnerable: as soon as one such component is missing, they are susceptible to losing the thread, and need to explore all components anew. They easily panic with incomplete, missing elements. Yet, the processing style does have its advantages: by its enhanced attention to detail, the autistic person is less easily distracted by the surrounding structures, enabling it to discern the individual component parts better (Shah & Frith, 1983). This cognitive style has comprehensively been investigated in the visuoperceptual domain in particular (see Simmons, Robertson, McKay, Toal, McAleer, & Pollick, 2009), and only little research had been done on visuomotor functioning. Moreover, most of these studies focused on children with the severest type of ASD, with little being known about the much larger group of children with milder variants of ASD.

That a cognitive bias for local rather than global processing is also hindering children within the autism spectrum during the execution of motor movements is exactly what Cattaneo and colleagues found. In a series of studies (2007; 2009) on ASD, they observed a lack of motor anticipation for the subsequent action within a chain of multiple actions. Whereas typically developing children anticipated the next step within a motor chain, the children with ASD only activated the muscles directly involved in the ongoing action. In other words, their bias for a local strategy not only impairs their visuoperceptual performance but also the execution of visuomotor tasks.

The main research questions in the studies presented in this dissertation were: Is the visuoperceptual processing of children at the milder end of the autism spectrum different from that of typically developing children and is it indeed characterized by a preference for local over global processing as is known to be the case in children with severe ASD? Are the visuomotor processes distinct from those observed in typically developing children? And what is the role and effect of the supposed atypical visual perception on their motor functioning?

I. VISUAL PERCEPTION

In the next sections the research pertaining to the visual perception of children with 'mild ASD' (at the time denoted as the PDD group) as reported in this thesis is summarised. The question was whether children with PDD also display deficiencies in visuoperceptual processing as is described for children with 'severe ASD' (i.e. autism).

In our investigations we made use of two tasks: the Embedded Figures Test (EFT), and a visual completion task. In the studies featuring the EFT (Chapters 2 and 4), we examined whether, similar to children with severe ASD (autism), children with mild ASD (PDD) also have a detail-oriented cognitive style. Broadly speaking, the central coherence theory states that when coherence is poor, this is caused by deficient processes in rendering context and meaning. As a result, children with ASD are less hindered by the surrounding context or meaning and pay preferential attention to the constituent details and parts: their cognition is characterized by a detail-oriented style.

In the study using the visual completion task (Chapter 5) we looked into the children's visuoperceptual performance at a more basic level where higher-order cognitive processes play no or a lesser role. The main purpose was to investigate whether also at this lower level differences could be found between children with PDD and age-matched controls.

a. Embedded Figures Tests

In 1983 Shah and Frith talked of an "islet of ability" in children with autism: better than their typically developing peers, the young children with autism were able to point out shapes and forms that were hidden (embedded) in a larger, meaningful figure. The authors posited that in their visual perception the autistic children were less hindered by the connotations of the overall image than the controls. The study described in Chapter 2 expands on this observation; we tried to replicate Frith's experiment with the Children's Embedded Figures Test (CEFT) but now in children with milder ASD (PDD), typical developers (TD), and another clinical group, i.e. children with Tourette Syndrome (TS). Apart from being less severely affected than Shah and Frith's children at the functional level, our boys with PDD were older and cognitively more advanced. The results of our trials did not meet our expectations: the children in the PDD group were not superior to the controls in detecting the hidden shapes: the three groups were similarly accurate and fast. The task seemed too simple, lacking the complexity and sensitivity to make out any differences in performance.

In a second experiment (Chapter 4) we only presented the more complex items of the CEFT, added an even more difficult version of the task, the adult Embedded Figures Test (EFT), and also included children with ASD whose cognitive and behavioural functioning was more severely affected. Again, the CEFT items failed to uncover any differences between the various groups (percentage correct ASD: 91%; controls: 87%). Even the group with more severe ASD did not perform better than the other groups. Not so for the EFT: the performance of the combined ASD group was superior to that of the control group (56% vs 48% correct trials), with performance times being similar. Within the ASD group, and corrected for age and IQ, the children with more severe ASD symptoms scored better (60.4% correct trials) than their peers with milder symptoms (55% correct) and the controls (46.8% correct).

In other words, although subtly but unmistakably so, children with ASD do show superior skills at spotting embedded figures in a larger context provided the complexity and sensitivity of the test is sufficiently high to avoid general ceiling effects. Corrected for age and IQ, children with more severe ASD are indeed better at detecting embedded figures than their typically developing peers. With the boys with milder ASD (PDD) performing at an intermediate level, also this latter group appears to be less diverted by contextual elements, which coincides with the notion that children with autism are less perceptive of global structures and better at perceiving details.

b. Visual completion task

Our findings so far suggested that detail-oriented visuoperceptual processing distinguishes children with ASD from typically developing children. However, the cognitive processes tested with the EFT are of a relatively high order where, in addition to the more fundamental perceptual processes, memory, i.e. recognition of previously perceived shapes, and assigning meaning play a role.

In contrast, the perceptual process of visual completion (see Chapter 5) involves lower-order perceptual processes. At four months of age, our visual system already 'sees' complete objects even when the objects are in fact only partly visible (Kellman & Spelke, 1983). A circle partially hidden from view by a superimposed rectangle simply looks to us like a full circle. In our perception, partly occluded objects are transformed by the visual system into sensations of more complete objects, a cognitive process that is referred to as visual completion. In our everyday lives we continuously come across objects that are partially occluded and we still perceive them as complete and not as fragmented. In the study reported in Chapter 5 we investigated whether the more detail-oriented visual processing in children with milder ASD symptoms (PDD) at a lower level would be reflected in a lesser degree of integration (visual completion). Would children with PDD show comparable visual completion skills to those exhibited by typical developers (TD) or would we find evidence of diminished contextual abilities complicating the visual completion process in the ASD group? At the time, no study had examined visual completion in children, let alone children with ASD.

How this completion process evolves is dependent on the properties of the stimuli (object), such as their degree of regularity. Two completion processes are distinguished. If the perception of a shape is based on the information at the location where the partly occluded shape meets the occluding superimposed figure, resulting in a linear or curved continuation of the occluded contours, (Kellman & Shipley, 1991; Fantoni & Gerbino, 2003), it is referred to as 'local completion'. If, however, it is based on more contextual information from the occluded shape, e.g. symmetry or iteration (Van Lier, van der Helm, & Leeuwenberg, 1995; Van Lier, 1999), it is referred to as global completion (see Chapter 5, Figure 5.2). It is generally agreed that certain properties of the stimuli, such as the type of regularity and the extent to which it is occluded, determine how the presented shape is perceived, i.e. the manner in which it is completed (Sekuler, 1994; Van Lier, van der Helm, & Leeuwenberg, 1995; Van Lier & Wagemans, 1999). Therefore, our question should rather read: Would children with PDD show a tendency towards a local-completion style or would they, similarly to what healthy adults do (De Wit & Van Lier, 2002), utilise the structure and regularity of the occluded figure and thus adopt a global-completion strategy (see Chapter 5, Figure 5.2)?

The study described in Chapter 5 comprised two experiments. In the first, we looked whether children with PDD (N = 19) similarly to typically developing peers (N = 28), would perceive simple, partly occluded figures as their wholes. Here, we used the primed-matching paradigm (Beller, 1971; Sekuler & Palmer, 1992; De Wit & Van Lier, 2002), which makes it possible to implicitly test how different images are perceived. In the task, the participants need to decide as quickly as possible (by pressing either a 'match' or a 'no-match' button) whether two adjacently presented figures (i.e. the test pair) are identical or different from each other. Showing a similar prime before the test pair speeds up the match decision. This facilitation, the priming effect, depends on the perceived similarity between the prime and the two shapes in the test pairs. This would enable us to determine whether the children perceived the occluded shapes as a whole figure or not. The results showed that both groups evidenced priming effects of occluded and full primes on full test pairs (circles), indicating that visual completion processes proceeded equally well in both groups. This implied that the children with PDD had managed to integrate partial visual information into a whole equally well as the controls. However, this similarity between the groups only held for the simple shapes (circles). With the more complex mosaic figures, the priming effect was far smaller in the PDD group than it was in the control group.

In the second experiment we presented even more complex figures, such as many-pointed stars. Would the children with PDD now show a diminished contextual completion, i.e. a tendency towards a local-completion style or would they, similarly to what healthy adults do (De Wit & Van Lier, 2002), utilise the structure and regularity of a figure and thus adopt a global-completion strategy (see Chapter 5, Figure 5.2)? The results showed that in the control group, global and occluded primes generated a priming effect on global test pairs and not on local test pairs. A similar pattern was found in the PDD group for global and occluded primes. Both groups, then, showed global completion, i.e. both had utilised contextual information.

Notwithstanding the similarities, the results also revealed three differences between the groups. In the control group the priming effect was about equal for global and occluded primes on global test pairs, but in the PDD group the effect for the occluded primes on global test pairs was less strong than it was in the control group. Second, the control group showed a priming effect for local primes on local test pairs, whereas the PDD group almost did not. Third, in the control group the local primes strongly inhibited priming effects on global test pairs; in the PDD group this was barely the case. By contrast, in the control group the global primes hardly "inhibited" priming effects for the local test pairs; in the PDD groups this was strongly the case.

Accordingly, although we found evidence of a priming effect and visual completion in the children with PDD, and although they showed global completion as well, in some aspects their completion process still differed from what we saw in the typically developing children.

In the children with PDD the more complex shapes (mosaic primes in experiment 1 and local primes in experiment 2) triggered the completion process less or differently than they did in the controls. Also, some of the more complex shapes appeared not to prime decisions in identical pairs (local primes in experiment 2).

Based on these outcomes, we can conclude that at the level of visual completion there are distinguishing differences in the visual perception of children with PDD and typical developers. Our results indicate that boys with PDD have more difficulty perceiving unusual/unfamiliar or complex shapes. Our study thus supported the assumption that even at a low level of perception children with PDD differ from typically developing children.

II VISUOMOTOR INTEGRATION

The atypical visual perception of children with milder ASD appears to be a factor in tasks involving the detection of shapes (EFT). Our visual completion study also underscored the unique way they perceive elements and contexts. The question remained whether this atypical style of visual information processing has an effect on the way they plan and coordinate their motor actions. We used three tasks to put this to the test: (1) the Rey Complex Figure Test (Rey's CFT), (2) a task assessing visual and non-visual tracking (in analogy to the paradigm used by Hermelin and O'Connor, 1970) and (3) the Developmental Test of Visual-Motor Integration (VMI; Beery, 1967, revised 1997). The study group consisted of 12 boys with PDD with 12 typically developing boys and 12 boys with Tourette Syndrome (TS) participating as controls.

a. Rey's Complex Figure Test

In Chapter 2 we probed whether copying and reproducing a complex picture would reveal a more detail-oriented cognitive style in children with milder symptoms of ASD (PDD) as we assumed. In Rey's CFT, a graphic figure (Chapter 2, Figure 2.2) needs to be copied and later drawn from memory both in a recall and a delayed recall condition. For our data analyses we adhered to the elaborated scoring system of Waber and Holmes (1985; 1986), which provides an organisation score reflecting structural completeness, a style score denoting a configurative, a detail-oriented, or an intermediate drawing strategy, an accuracy score, and an error score (rotations, misplacements, perseverations and conflations). Within the figure itself, a distinction is made between structural and incidental elements to evaluate global and local information processing. To have an additional measure of drawing style, we added a fragmentation score reflecting the number of strokes that were used to construct the structural elements (i.e. base rectangle and intersecting lines; Chapter 2, Figure 2.3).

Compared to the two comparison groups (TD and TS) and especially in the recall conditions but also in the simpler copy condition, the boys with PDD achieved lower organisation and higher detail-oriented style scores. The TS and TD groups succeeded in correctly reproducing over 90% of the structural elements in both recall conditions where, with scores under 76%, the PDD groups performed significantly poorer. All three groups succeeded in reproducing about 75% of the incidental elements in the recall conditions. The boys with PDD did not discriminate between the two types of elements; they appear to view the structural elements as separate entities without any internal coherence. To them, a rectangle consists of four separate elements rather than together constitute a single (structural) entity. They inevitably drew these elements as they drew incidental elements, accounting for their relatively low scores (75% correct). Their way of drawing thus confirmed our expectation that children with PDD employ a detail-oriented perceptual style that leads to a fragmented reproduction of Rey's figure. Unlike the children in the two control groups, they were probably unable to derive support from the structures and context of the target figure. Compared to the controls (TS: 4.0 %; TD: 3.0%), they also produced more errors (10.6%), most notably in the delayed recall condition. Moreover, their fragmentation scores showed that, from the copy condition onwards, the PDD boys used more line segments to draw the elements than their peers in the other two groups, with scores remaining high in both recall conditions.

The results of our study clearly demonstrated that in drawing a complex figure boys with PDD use a more piecemeal strategy than age peers, making less use of (or perhaps not perceiving its) context and structures. This processing style goes hand in hand with a poor performance outcome (reproduction) and a quantitative and qualitative loss of information. We hence inferred that the performance differences were mainly attributable to differences in visuoperceptual processing because in the more simple copying condition we already observed minor but statistically significant differences in organisation (completeness), drawing style, and fragmentation, a condition in which any planning and memory problems can be easily remediated by inspecting the target figure.

b. Tracking task

To further understand the effect of visual perception on motor action and in view of the fact that the study in which Hermelin and O'Connor (1970) had children with autism trace a visible or masked track had never been replicated, we decided to also ask our study group to complete a tracking task (Chapter 3). In our replication study we administered a slightly modified version of their original task. In a blinded and an unblinded condition, the boys with PDD, Tourette's and typically developing peers were instructed to trace preprinted grooves in a perspex plate with a non-inking electronic pen. They completed four different tracks increasing in complexity. By manipulating the visibility of the tracks, we were able to investigate the effects of visual perception on the children's motor performance. Please note that speed is the only dependent variable in this task, because grooved tracks do not allow for gross inaccurate movements, while pushing the pen in an improper direction reduces movement speed.

Like Hermelin and O'Connor (1970) in their group of autistic children, we found that the boys with PDD did not trace the tracks any faster in the vision condition than the combined control group (9.17s vs 8.30s), while in the blinded condition they did, and much faster so (16.3s vs 23.7s). Looking into the effect of the presentation order of the conditions, we found that the controls had benefited by having seen the tracks in the previous unblinded condition: they performed faster in the blinded condition. This, however, was not the case for the PDD group. The performance of the controls strongly improved in the first three unblinded trials, most markedly if they had not seen the tracks earlier (blinded first: 21s vs 9s; unblinded first: 12s vs 8s). This effect was far less strong in the PDD group: having visual control of the tracks only marginally improved their performance (blinded first, 8s vs unblinded 7s; unblinded first: 12s vs 11s). Most notable was the finding that the boys with PDD that had seen the tracks in the first condition tracked more slowly in both conditions than their peers with PDD who had not. This was contrary to what we observed in the controls: they performed much better in the blinded condition if they had seen the track before, even in the fourth and most complex track (blinded first: 54s vs 34s). The PDD group's performance improved to a far lesser extent (39s vs 32s). It was also striking that it was only in tracing the blinded tracks that they showed their superior performance in velocity; in the unblinded trials no group differences were observed.

Apparently, in tracking children with PDD rely far less on visual cues to plan or correct their motor actions. Arguably, for their remarkable tracking performance haptic information seems to suffice, rendering visual information largely expendable.

c. The Beery-Buktenica Developmental Test of Visual-Motor Integration (VMI)

The third task we used to evaluate the visuomotor performance of our three groups (PDD, Tourette's, typical developers) was the Beery's VMI (1997). In this task children are asked to copy (motor integration), compare (visual perception) and trace (motor coordination) geometric figures of increasing complexity. We compared the performance of the three groups in terms of their standard scores, their visuoperceptual performance scores and motor execution times (see also Chapter 3).

As the results recorded for the two control groups were statistically comparable for all three scales, the groups were pooled. The VMI total scores for the boys with PDD were significantly lower, i.e. their visuomotor integration was decidedly poorer than that of the pooled control group. Contrary to our expectations, we found no such performance differences on the visual perception or motor coordination items. Another noteworthy finding was that the PDD boys were faster in drawing than the controls (correct trials only) irrespective of the complexity of the stimulus figures (i.e. even when copying simple lines: 3.8 vs 2.9 cm/s).

Although the study was not designed to investigate the mechanisms underlying the high drawing velocities the boys with PDD achieved, observing their behaviour led us to assume that in PDD visuomotor control is limited in that they rely less on visual information to monitor their movements, which is in agreement with what we had earlier observed in our tracking task. This limited use of visual information can save them time and enhance their motor speed.

An explanation for their lower VMI total scores is not unequivocal. In figure copying various processes are at play: the visual perception of the target figure, the formation of a mental representation and its storage into working memory, visuospatial planning, motor planning, motor execution, monitoring, and motor adjustments guided by the graphic reproduction (see also Mottron, Belleville, & Ménard, 1999). Although each of these processes may be implicated, a poor VMI score may also result from an atypical processing of visual information. If this processing style entails a distinct focus on details rather than on the entirety of the figure, the formation of the representation in memory of the figure that has to be copied will be hampered.

IN SUMMARY

The main research questions driving the studies presented in this dissertation were: (1) Are visuoperceptual processes deficient in children at the milder end of the autism spectrum in a way that is similar as is described for children with severe autism and different from typically developing children; and, (2) If so, what are the effects of such an atypical visual perception on their motor functioning?

With respect to the first question, this is what we found:

The differences we observed between the children with milder and more severe ASD (Chapter 4) were only quantitative: in the EFT the disembedding capacity of the whole ASD group was better than it was for the control group. Looking more closely, the children with milder ASD (PDD) performed in between the children with severe ASD (autism) and the controls. We also found evidence of atypical visuoperceptual processing with Rey's CFT (Chapter 2): the milder ASD (PDD) group showed detail-oriented processing. The tracking task study (Chapter 3) revealed a lesser use of visual perception in PDD, a result that Hermelin and O'Connor had reported for children with autism in 1970. As to the nature of these atypical characteristics of visual processing in PDD, it shared the processing style that was earlier reported for children with autism while we found no qualitative differences between the two populations.

Moreover, at the lower level of visual perception (Chapter 5), the processing of complex shapes also appears to be deficient in mild ASD and different from typically developing children.

As to the second question, we found that in boys with mild ASD the effect of visual information on motor action was various. With Rey's CFT (Chapter 2) and the VMI (Chapter 3) we saw that the immediate reproduction (copying) of the figures was poor while delayed reproduction of Rey's Figure led to a substantial loss of quantitative and qualitative content. In the tracking task (Chapter 3) visual guidance appeared diminished as compared to the role it played in typically developing peers: the boys with mild ASD seemed to rely less on the available visuoperceptual information to plan and monitor their motor actions, which speeded up their motor performance.

The results in all the studies presented in this thesis suggest that it is the visual processing deficiencies that seem a relevant element in the visuomotor peculiarities common in children with ASD. This "atypical" visual perception not only plays a role in high-level cognitive processes but, given their deviant visual completion performance, also characterises their functioning at the lower level.

THEORETICAL IMPLICATIONS

The findings described in this dissertation contribute to the existing knowledge and theories on the mechanisms underlying ASD.

I. The perceptual divergences we observed in our groups with mild and more severe ASD point to atypical/ deficient visual information processing that is characterised by a detail-oriented cognitive style, a style first described and denoted as *weak (central) coherence* by Frith (1989) and Happé and Frith (1996; 2006). In 1989 Uta Frith posited that "In the normal cognitive system there is a built-in propensity to form coherence over as wide a range of stimuli as possible, and to generalize over as wide a range of contexts as possible. It is this drive that results in grand systems of thought, and it is this capacity for coherence that is diminished in children with autism." In a thorough review and with their own study, White and Soldaña (2011) sowed doubts about the evidence of the central coherence theory that was gained with embedded figures tests. Both in their review and their own study they underscored that children with ASD did not show superior disembedding capacities, a skill frequently used to demonstrate weak central coherence. Yet, in contrast to their findings and conclusions, our study (Chapter 4) showed how embedding tests can be used to demonstrate weak coherence: if the sensitivity of the test is taken into account in that the task is sufficiently difficult to challenge the control group, performance differences do become apparent.

6

One leading theory within the weak central coherence account is the enhanced perceptual functioning model developed by Mottron and Burack (2001) and their group, investigating functioning of 'savants' and non-retarded people with autism in visual and auditory perceptual cognitive tasks and by brain imaging. They assert that the detail-oriented perception of children with ASD results from an enhancement of low-level (auditory and) visual processing leading to an attentional focus on low-level sensory phenomena and accordingly propose the existence of an initial deficit in the recognition of complex patterns followed by compensatory increases in aspects of functioning ("as an obsessive search for individual – constituent (red.) – lines", 2001, p143), which results in an enhanced perception of the components of these complex patterns. The default setting, they suppose, of "autistic" perception is more locally oriented than that of typically developing individuals (Mottron, Dawson, Soulières, Hubert, & Burack, 2006). They disagree with Happé's interpretation of cognitive styles as being facultative in nature and point to differences in cognition at fairly low levels in which this style is inevitable and cannot be consciously selected. The results on the EFT we obtained (Chapter 4) are in line with this account. In addition, our findings on the completion task (Chapter 5) may also reflect such a deficient recognition of complex patterns at a lower level. However, we did not find any enhanced perception on Rey's CFT (Chapter 2): compared to the control groups the boys with PDD did not reproduce more incidental elements. Rather, their reproduction of structural elements was affected, suggesting a reduced perception and generation of global structures at a higher visual-cognitive level.

Researching the cognitive strengths and superior discrimination abilities of individuals with autism, Plaisted (2001) formulated a model of reduced generalisation as an alternative for the weak central coherence account. She reasons that "differences in the way the autistic brain processes how similar and how different things are to one another" lead to a reduced generalisation capacity in children with ASD. Our combined results, including the deficient priming and completion processes for more complex shapes, go beyond the enhanced perceptual function theory and support Happé (1996) and Plaisted, Saksida, Alcantara, and Weisblatt (2003) in their suggestion of the existence of more fundamental perceptual deficits in children with ASD, underscoring the findings of many visuoperception studies in individuals with ASD as reviewed by Simmons and colleagues (2009).

II. Alternatively and seen from the perspective of the executive function theory (Russell, 1997), the results we obtained with Rey's CFT, the VMI and our tracking task (Chapter 3) might point to problems in planning and guiding motor actions. According to this theory, in children with autism there is "a severe early disruption in planning of complex behaviour, due to a severe deficit in working memory" (Pennington, Rogers, Benetto, McMahon Griffith, Reed, & Shyu, 1997, p148). Executive functions encompass different domains such as response inhibition, working memory, cognitive flexibility (set shifting), planning, and fluency (Geurts, Verté, Oosterlaan, Roeyers, & Sergeant, 2004). However, the performance results on the EFT and particularly those on the visual completion task clearly suggest lower-level perceptual problems. In completion tasks no executive strategies such as inhibition or task switching are involved; the tasks simply gauge how perception is organised. What matters are the interpretation of the retinal image and the creation of stable and reliable extrapolations and representations. These processes define working memory processes and planning: representations govern planning and unburden the working memory. On the other hand, it is quite possible to understand how planning and goal directing is hampered if perceptual processing and representation forming is impeded. Today, it is generally accepted that it is unlikely that executive dysfunctioning plays a primary causal role in autism (Griffith, Pennington, Wehner, & Rogers, 1999; Yerys, Hepburn, Pennington, & Rogers, 2007), but in an extensive review Pellicano (2012) demonstrates that it still plays a substantial role in the developmental outcomes of these children. Perceptual deficiencies draw heavily on working memory and planning capacities and lead to the development of compensating strategies frequently inhibiting natural pathways.

RECENT DEVELOPMENTS

From the 1980s until recent years, three major accounts on autism played a dominant role: theories of impaired social cognition, of impaired executive functioning and of weak coherence (see Chapter 1), including the younger enhanced perceptual function theory (Mottron & Burack, 2001). Still, none of these accounts could fully explain all the symptoms typically seen in ASD, nor is weak coherence reducible to executive dysfunctions, while deficits in social cognition seem independent of weak coherence (Happé & Frith, 2006). Recently, Lawson, Rees and Friston (2014) formulated a unifying hypothesis on autism, based on the predicting coding qualities of the brain. From the perspective of the predictive coding account (Picard & Friston, 2014), our brain is a "fantastic" organ generating expectations to explain our sensations. It is described as a predictive machine that continuously seeks to improve its beliefs through trial and error. The brain makes real-time predictions in different sensory streams and tests these against the sensory data it actually samples; it is an active organ of inference, constantly predicting its sensory input. By doing so, models are generated of likely prospective sensations, generative models based on experience, to enable perceptual inference or synthesis. In this theory, predictions (priors) and prediction errors are precision-weighted, leading to different degrees of confidence. The brain not only predicts the content of the sensory input but also predicts the precision or degree of confidence that contextualises this input (Picard & Friston, 2014). Predictive processing is not limited to sensory input alone, it is supposed to also apply to action. Here, the brain is thought to generate continuous predictions about the estimated location of the limbs and eyes consistent with the goal of the upcoming movement.

Pellicano and Burr (2012) proposed to conceive the sensory and non-social symptoms of autism as a deviation in the interpretation of sensory input that yields percepts. As compared to typically developing people, the perceptual experience of people with autism would be less modulated by prior beliefs, they suggested, because their "priors" would be “attenuated”. Lawson, Rees and Friston (2014) hypothesised that aberrant encoding of the precision accounts for the social and non-social peculiarities in autism, because of its role in coordinating the dynamics of perception, action and social behaviour, and may thus also underlie other neuropsychiatric syndromes.

According to this youngest account, visual completion might be seen as an inference of an object before this object is completely disclosed, based on a sufficient match between the prediction and a partial visual cue. The results of our study on visual completion (Chapter 5) suggest that more complex forms are processed less efficiently by children with PDD as compared to typically developing youngsters: the priming effect is less strong and the completion attenuated (Chapter 5, Figures 5.4 and 5.5). Consistent with the aberrant precision account of autism (APA; Lawson, Rees, & Friston, 2014), these results might be conceived as the consequence of an aberrance in precision with regard to the prediction for these figures (mosaic form and local form), where the precision in the prediction of the simple forms (circle and global form) is comparable to that of healthy peers. The impression exists that as a consequence of this aberrant precision-weighting the formation of a representation of more complex shapes is hindered or delayed. Yet, since the APA account has only recently been formulated, more research is required to test the consequences of this new, unifying account and the relationship to ASD. Differences in the development of confidence in prediction between children with ASD and typical developers might then be studied in visual completion tasks but also in more dynamic visuomotor paradigms.

IN CONCLUSION

With the differences we observed being mostly quantitative and not qualitative in nature, the results we obtained with the studies presented in this thesis strongly suggest that the processes that underlie the impairments in children with milder and more severe symptoms of ASD are similar. These findings then justify the practice to offer all children with disorders within the autism spectrum similar treatment and management schemes, which notion has been generally acknowledged with the introduction of the DSM 5.

The effects of the deviant visual processing we found influence the visual motor functioning in an atypical and divers way. This visual processing not only differs from the processing in typically developing children at a high cognitive level, but also at a lower more basic level. Their reduced ability to quickly “see” a global structure implies that children with ASD live in and have to learn to deal with a chaotic environment, which requires a straightforward, explanatory, empowering approach of professionals and others directly involved in these children's lives, as well as an adequately informed external support system. Awareness of how they have a tendency to go about things in a piecemeal, detail-oriented fashion will not only help the children themselves to cope better with their ASD and their parents to understand and come to grips with their child's weaknesses, it will also enable both children and parents to put their strengths to good use in their daily lives. Understanding the uncertainty of their judgements about the (in)variability of their surroundings may help us, professionals and informal caregivers alike, devise more effective ways to help these children participate (more) easily in today's fast and challenging world from an early age onwards.

The results we obtained in our group of children with (mild) ASD using the VMI and tracking tasks point to serious deficits in visuomotor integration and warrant replication. Among possible causes, such a visuomotor deficit might originate from a deficiency in visual processing, which might also hamper the visual support of motor activities. Visual guidance is not only needed to provide feedback during motor execution but is of particular importance in the early stages of motor learning when new movement patterns have to be acquired. Another potential cause for a visuomotor deficit might be found in the fragmentation of motor programmes. In ASD, the tendency of local processing seems not to be restricted to visual perception alone and may also present in other perceptual domains (auditory, haptic and proprioceptive) (see Molesworth, Chevallier, Happé, & Hampton, 2015) and might even appear in language and executive functioning (Pellicano, 2012; Geurts, Verté, Oosterlaan, Roeyers, & Sergeant, 2004). It is therefore quite possible that also motor programming is affected in the sense that in ASD motor planning is less global, i.e. more restricted to the execution of ongoing movements while upcoming actions are less or not anticipated (Cattaneo et al., 2007; Fabbri-Destro, Cattaneo, Boria, S., & Rizzolatti, 2009).

In our research, we did not investigate the potential contribution of fragmented motor execution to the observed deficit in visuomotor integration. Our studies were targeted at a higher phenomenal level with the aim to explore a rather neglected area in ASD research and to demonstrate that visuomotor performance might be deficient in this continuum of disorders. Unravelling the disturbances in the underlying processes would be a fruitful next step.

If our findings on visual completion are replicated, visual perception in ASD might also be examined in younger children to better understand the (typical) development of this deviant processing and its implications. Such studies might then foster research to help us identify and diagnose ASD at a younger age. It is, moreover, of great interest to explore how the brain creates object representations to recognise objects. Is this process deficient in children with ASD or is it partly delayed, with the capacity developing at an older age? Knowing which aspects in their perception are delayed, deficient or deviant, and to understand to what developmental deviations, compensating strategies and coping styles this leads, will enable us to support all children with ASD better in their daily lives and prevent us from asking too much of them and to guide them to interact with the world around them in an optimal manner.

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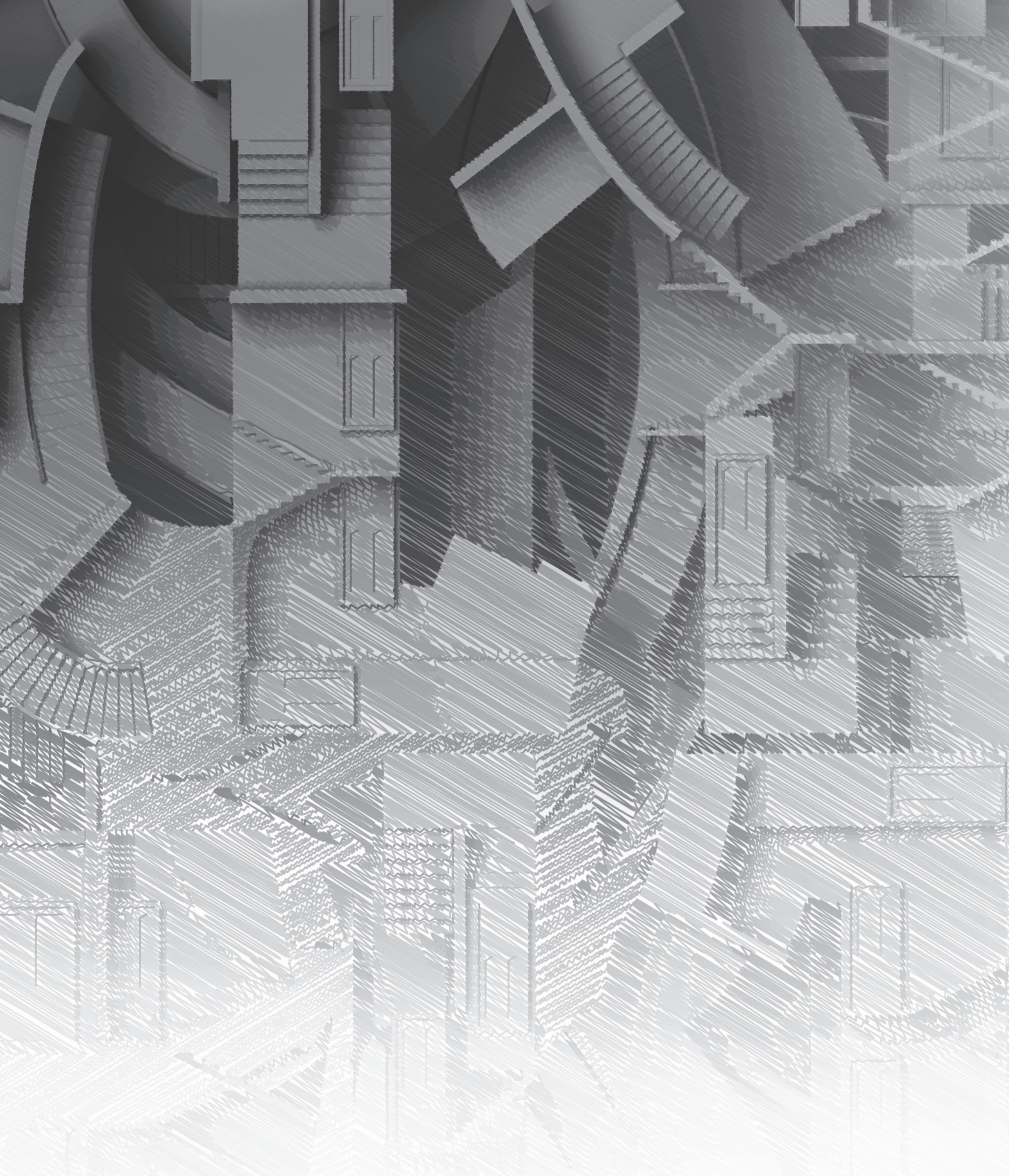


Summary

Drawing out the visuomotor abilities of children with mild autism spectrum disorder

Children with autism spectrum disorder (ASD) have major and pervasive problems communicating and interacting socially and show restricted, repetitive patterns of behaviour, interests or activities. Many domains of functioning may be affected, not only by the ability to initiate and maintain social contacts, but also speech and language development, planning and organisational capacity, perception, and often also motor skills.

The research presented in this thesis was inspired by the ample observations that children with ASD take longer to learn to ride a bike or swim and tend to lag behind in learning to write. Although deviations in motor skills are not part of the criteria for an ASD diagnosis, the children's motor development does show typical tendencies. Clinical observations suggest that children with ASD have more problems drawing a human figure than their typically developing age peers (Chapter 1). Accordingly, the main focus of this thesis was to gain a better understanding of the visuomotor functioning of children diagnosed with a disorder in the autism spectrum.

A second priority of the present research was to not zoom in on children with the severest ASD type, i.e. those suffering from autistic disorder (AD), but on the far larger group (Chapter 1) diagnosed with milder variants of ASD formerly classified as Asperger syndrome or Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS), together referred to as PDD in some chapters of this thesis.

The observed difficulties in drawing may not solely be due to a motor problem but could also or alternatively result from biases or deficits in the way children with ASD perceive and conceptualise the world around them. One of the three guiding neurocognitive theories on the underlying mechanisms of autism is the central coherence hypothesis formulated by Frith in 1989 and elaborated by Happé and Frith in 1996 and 2006. Their theory proposes that people with autism show a strong tendency to focus on details and that they make little use of the context or existing structures in which component parts are embedded: they are masters in detail but have little eye for overall structures. In 1999 Francesca Happé speaks of "a cognitive style biased towards local rather than global information processing".

With the studies described in this dissertation we specifically sought to learn whether in children diagnosed with disorders at the milder end of the autism spectrum any deficits in visuoperceptual processes would be similar to those described for children with severe autism, how any atypical visual perception would affect their motor functioning, and how their performance would deviate from that observed in typically developing children. Chapters 2 to 5 featured four separate studies that addressed these questions. Below, the results of these studies are summarised under two headings: visual perception and visuomotor integration.

I. VISUAL PERCEPTION

To investigate visual perception we utilized two tasks: the Embedded Figures Test (EFT) and a visual completion task. In the studies featuring the EFT (Chapters 2 and 4), we examined whether, similar to children with severe ASD (autism), children with mild ASD (PDD) also have a detail-oriented cognitive style. In the study using the visual completion task (Chapter 5) we looked into the children's visuoperceptual performance at a more basic level where higher-order cognitive processes play no or a lesser role.

a. Embedded Figures Tests

In 1983 Shah and Frith had young children with autism complete the Children's Embedded Figures Test (CEFT) and found that they were far better at detecting forms that were hidden (embedded) in the larger, meaningful figure than their typically developing peers, which prompted them to coin the phrase "islet of ability" to characterise this special aptitude. The study described in Chapter 2 expands on this observation in that we tried to replicate Frith's experiment with the CEFT but now in children with milder ASD (PDD), another clinical group, i.e. children with Tourette Syndrome (TS), and typical developers (TD). Contrary to expectations, we found that the children in the PDD group were not superior in detecting the hidden shapes: the three groups were similarly fast and accurate, which made us conclude that the task was too simple and lacked the complexity and sensitivity to detect subtle differences in performance.

In a second experiment (Chapter 4) we added a more difficult version of the task, the adult Embedded Figures Test (EFT), and also included children with ASD whose cognitive and behavioural functioning was more severely affected. Again, the CEFT failed to uncover any group differences but the EFT did: the performance of the combined ASD group was superior to that of the control group (56% versus 48% correct trials), while performance times were similar. Within the ASD group, and corrected for age and IQ, the children with more severe ASD symptoms scored better (60% correct trials) than their peers with milder symptoms (55% correct) and the controls (47% correct).

In short, although subtly, the children with ASD did show superior skills at spotting embedded figures once general ceiling effects were precluded by offering them a more complex task. Even the boys with milder ASD (PDD) appeared to be less distracted by contextual elements, coinciding with the view that children with autism are less perceptive of global structures and better at perceiving details.

b. Visual completion task

The cognitive abilities tested with the EFT are of a relatively high order, whereas visual completion tasks (see Chapter 5) gauge lower-order perceptual processes. Even at four months of age, our visual system already 'sees' complete objects even when the objects are in fact only partly visible: it 'perfects' our perceptions by transmuting partially occluded objects into their more complete forms, which mental process is referred to as visual completion. In the study reported in Chapter 5 we investigated whether at this lower perceptual level the more detail-oriented visual processing style we observed in our children with milder ASD symptoms (PDD) would be reflected in a lesser degree of integration as compared to that seen in typical developers (TD). At the time of our study, visual completion had not yet been examined in any child population.

Building on earlier research in adult populations, we used the primed-matching paradigm to implicitly test how forms are perceived, exploiting the "priming effect" (PE), where it is assumed that by first showing a target shape (prime), a subsequent identical shape will be recognised more quickly. This effect is also found when a pair of shapes that is identical to the priming shape is presented. By manipulating the forms, i.e. by also offering partly occluded or deviating shapes, we hoped to identify differences in lower-level visual perception.

The study described in Chapter 5 comprised two experiments. The results of the first (see Figure 5.4) showed that both the PDD and the TD group showed PEs following occluded and full primes on full test pairs (circles), indicating that as far as simple shapes were concerned visual completion processes proceeded equally well in both groups. With the more complex mosaic figures, however, PEs were far smaller in the PDD group than they were in the control group.

The results of the second experiment with even more complex figures (see Figure 5.5) showed similar group-specific patterns for global and occluded primes. Again, both groups showed global completion, i.e. all children had utilised contextual information, while the groups differed in three specific aspects: in the TD group the PEs for global and occluded primes on global test pairs were about equal, whereas in the PDD group the effects for the occluded primes on global test pairs were less strong than they were in the control group. Second, the controls showed PEs for local primes on local test pairs, whereas the boys with PDD hardly did so. Third, in the control group the local primes strongly reduced PEs to global test pairs, where in the PDD group this was barely the case. By contrast, in the control group the global primes hardly "reduced" PEs for the local test pairs, while in the PDD groups they strongly did so.

Our two visual-completion tasks showed that the boys with PDD indeed had more difficulty perceiving unusual/unfamiliar or complex shapes than their typically developing peers, prompting us to conclude that also low-level visual perceptual processing is affected in mild ASD.

II VISUOMOTOR INTEGRATION

Still needing to establish whether their atypical style of visual information processing also exerted an effect on the way boys with mild ASD plan and coordinate their motor actions, we presented 12 boys with PDD, 12 boys with Tourette Syndrome (TS) and 12 typically developing boys (TD) with three tasks: (1) the Rey Complex Figure Test (Rey's CFT), (2) a tracking task and (3) the Developmental Test of Visual-Motor Integration (VMI).

a. Rey's Complex Figure Test

In Chapter 2 we investigated whether copying and reproducing a complex picture (Rey's Complex Figure; see Figure 2.2) would also uncover a more detail-oriented cognitive style in boys with PDD. Compared to the two comparison groups, the boys with PDD achieved lower organisation and higher detail-oriented style scores even in the simple copy condition but most prominently so in the recall conditions (Figures 2.3 and 2.4). All three groups succeeded in reproducing about 75% of the incidental elements in the recall conditions. The TS and TD groups scored better on the structural elements in the recall conditions, where the boys with PDD did not discriminate between structural and incidental elements; they appeared to view the structural elements as separate entities without any internal coherence.

The results of this study clearly underscored that in drawing a complex figure the boys with PDD used a more piecemeal strategy than their age peers while they made less use of (or perhaps not perceiving its) context and structures.

b. Tracking task

Since the study in which Hermelin and O'Connor (1970) had children with autism trace a visible or masked track had never been replicated, we had our study and control groups complete a slightly modified version of their original tracking task (Chapter 3) to delineate any differential effects of visuoperceptual processing on motor performance further. Using four different tracks of increasing complexity, the boys with PDD, Tourette's and typically developing peers needed to trace preprinted grooves in a perspex plate with a non-inking electronic pen in a blinded and an unblinded condition.

Similar to Hermelin and O'Connor's observations (1970) in autistic children, we found that the boys with PDD did not trace the tracks any faster in the vision condition than the combined control group, while in the blinded condition they did, and much faster so. Most remarkably and unlike the controls who performed much better in the blinded condition after having seen the track before, the boys with PDD that had seen the tracks in the first condition tracked more slowly in both conditions than their peers with PDD who had not. It was also striking that the boys with PDD only outperformed the controls on velocity when tracing the blinded tracks; the unblinded trials yielded no group differences.

During tracking, visual information appeared expendable for the boys with PDD; they largely seemed to be guided by haptic information, from which we inferred that children with PDD rely far less on visual cues to plan or correct their motor actions.

c. The Beery-Buktenica Developmental Test of Visual-Motor Integration (VMI)

We also had our three groups complete Beery's VMI in which test children are asked to copy (motor integration), compare (visual perception) and trace (motor coordination) geometric figures of increasing complexity (Chapter 3). The VMI total scores for the boys with PDD were significantly lower, i.e. their visuomotor integration was decidedly poorer than that of the two control groups. Surprisingly, we found no such performance differences for the visual perception or motor coordination items. If the processing style of children with PDD is indeed characterised by a distinct focus on details rather than on the figure as a whole, the formation of the representation in memory of the figure to be copied will be hampered, which may then account for their lower scores. We also noted that, compared to the times recorded for correct trials in the controls, drawing times were always faster in the PDD group, i.e. the complexity of the stimulus figures was of no consequence.

Although we never sought to explain the underlying mechanisms of the high drawing velocities in the boys with PDD as such, the results of our visual-motor integration experiment and the findings on our earlier tracking task did persuade us to infer that the role of visuomotor control is diminished in this group: boys with mild ASD appear to use visual information less than typical developers to monitor and adjust their movements, which saves them time, speeding up their motor actions.

DISCUSSION

Comparing the results of the four studies, in Chapter 6 we posed that our combined findings suggest that it is the visual processing deficiency that seems to be the most relevant factor in the visuomotor peculiarities that are so common in children with ASD and that it not only plays a role in higher-order cognitive processes but, given their deviant visual completion abilities, also affects basic, lower-order functions.

The divergences in perception we observed in our boys with mild and more severe ASD point to atypical or deficient visual information processing that is characterised by a detail-oriented cognitive style, a style Frith (1989) and Happé and Frith (1996; 2006) first uncovered and denoted as weak (central) coherence. Although in their comprehensive review and with their own study, White and Soldaña (2011) questioned the evidence of the central coherence theory gained with embedded figures tests, our studies (Chapter 4) showed that embedding tests can be used to demonstrate weak coherence provided their complexity is sufficiently high to challenge typical developers and to thus allow subtle but distinct performance differences to become apparent.

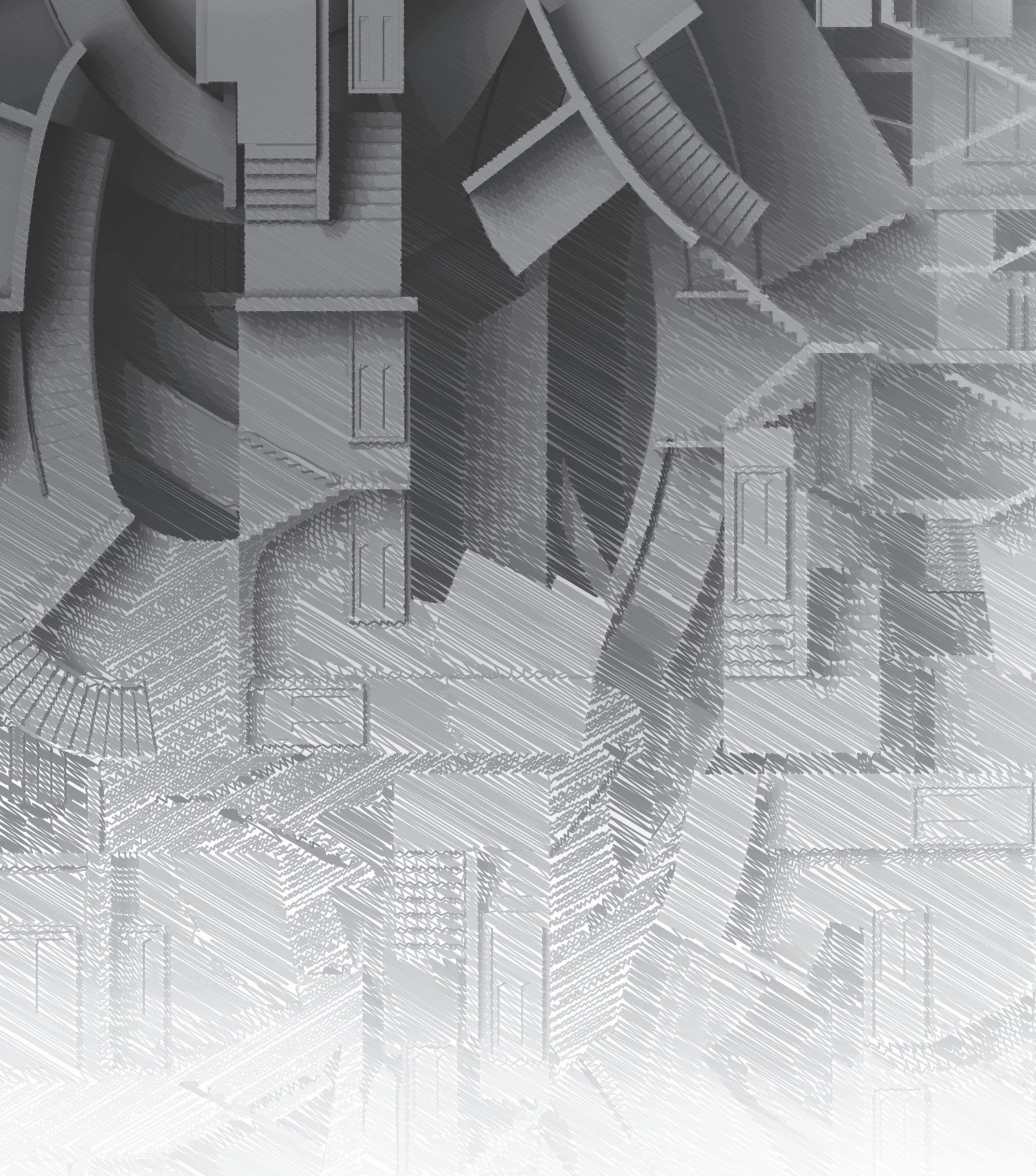
In their recent unifying hypothesis on autism that they based on the predicting coding qualities of the brain, Lawson, Rees, and Friston (2014) hypothesise that, because of its role in coordinating the dynamics of perception, action and social behaviour, it is an aberrant encoding of precision that accounts for the social and non-social peculiarities in autism and possibly also other neuropsychiatric syndromes. In the final chapter of this thesis we discussed our visual completion results in the context of this theory.

In conclusion

Since the group differences we obtained in the studies presented in this thesis were mostly quantitative, our combined results strongly suggest that the mechanisms that underlie the impairments in children with milder and more severe symptoms of ASD are similar in nature, validating the practice to offer all children with disorders within the autism spectrum similar treatment and management schemes, which notion is now generally acknowledged with the introduction of the DSM 5.

The children's deviant visual processing affects their visuomotor functioning in a very distinct way and at every functional level. It not only differs from how typically developing children process visual information at a high cognitive level, it also does so at the lower, more basic level.

Awareness that, like their peers with severe ASD, children with mild ASD have a tendency to go about things in a piecemeal, detail-oriented fashion will not only help them cope better with the consequences of their disorder, it will also enable their parents, (informal) caregivers and professionals to understand the child's weaknesses and foster its strengths. Knowing which aspects of their perception are delayed, deficient or deviant, and to understand to what developmental deviations, compensating strategies and coping styles this leads, will enable us to support children with ASD better in their daily lives. By providing them with individual guidance, we can help them optimise the way they interact with the world around them, improving their opportunities and quality of life.



Samenvatting

Een kenschets van de visuomotoriek bij kinderen met een milde autismespectrumstoornis

Kinderen met een autismespectrumstoornis (ASD) hebben ernstige problemen met de communicatie en sociale interactie, hetgeen ingrijpende gevolgen heeft voor het dagelijks functioneren. Tevens worden ze gekenmerkt door zich herhalende stereotiepe patronen in gedrag, interesses of activiteiten. Daarnaast kunnen veel functies zijn aangedaan, niet alleen het vermogen om sociale contacten aan te gaan en te onderhouden, maar ook de spraak- en taalontwikkeling, het planning- en organisatievermogen, de waarneming en ook vaak de motorische vaardigheden.

Het onderzoek in dit proefschrift is ingegeven door de klinische ervaring dat kinderen met ASD vaak laat leren fietsen of zwemmen en ook vaak moeilijk (goed) leren schrijven. Hoewel afwijkingen in motorische vaardigheid geen criterium zijn voor de diagnose ASD, laat de motorische ontwikkeling van deze kinderen dikwijls wel bijzonderheden zien. Uit observatie (beschreven in Hoofdstuk 1) kwam ook naar voren dat, in vergelijking met zich normaal ontwikkelende kinderen, kinderen met ASD moeite hebben met het tekenen van een mensfiguur. Het hoofddoel van dit proefschrift was dan ook een beter begrip te krijgen van de visuomotoriek van kinderen met ASD.

Een tweede doelstelling was niet zozeer in te zoomen op kinderen met autisme, de meest ernstige vorm van ASD, maar op de ongeveer driemaal grotere groep van kinderen met een mildere vorm van ASD, eerder omschreven als Aspergersyndroom en Pervasieve Ontwikkelingsstoornis-Niet Anderszins Omschreven (POS-NAO ofwel PDD-NOS), en in een aantal hoofdstukken van dit proefschrift aangeduid als ‘PDD’.

De beschreven problemen met tekenen hoeven niet (zuiver) het gevolg te zijn van een motorische problematiek, maar zouden ook (of vooral) kunnen voortkomen uit de wijze waarop kinderen met ASD de wereld waarnemen en begrijpen. Een van de drie richtinggevende neurocognitieve theorieën die de specifieke tekorten bij autisme trachten te verklaren is de centrale coherentiehypothese die Frith in 1989 formuleerde en die in 1996 en 2006 is aangevuld door Happé en Frith. De theorie veronderstelt dat mensen met autisme een sterke neiging hebben hun omgeving detailgericht waar te nemen, waarbij zij de context of omgevende structuren waar deze details deel van uitmaken weinig gebruiken. Ze zijn meester in het detail maar hebben weinig oog voor globale structuren. In 1999 spreekt Francesca Happé dan ook van ‘een cognitieve stijl die eerder neigt naar een lokale dan een globale wijze van informatie verwerken’.

Met de studies die in dit proefschrift beschreven worden wilden we nagaan in hoeverre kinderen met een milde vorm van ASD bijzonderheden in de visuele waarneming vertonen die vergelijkbaar zijn met die beschreven zijn bij kinderen met autisme, hoe die eventuele atypische visuele waarneming hun motorisch functioneren beïnvloedt en hoe hun visuomotoriek afwijkt van dat van kinderen die een standaard ontwikkeling vertonen.

Hoofdstuk 2 tot en met 5 omvatten vier studies waarin deze vragen nader zijn uitgewerkt. Hiernavolgend worden deze studies in twee rubrieken samengevat getiteld visuele perceptie en visuomotoriek.

I. VISUELE PERCEPTIE

In ons onderzoek naar visuele waarneming maakten we gebruik van twee taken: de Embedded Figures Test (EFT) en een zgn. visual-completion-taak. In de EFT-studies (Hoofdstuk 2 en 4) onderzochten we of kinderen met een milde vorm van ASD (PDD) net als kinderen met ernstig ASD (autisme) een detailgeoriënteerde cognitieve stijl hanteren. In de visual-completion-studie (Hoofdstuk 5) keken we naar een meer basaal niveau van waarnemen, waar cognitieve processen van hogere orde geen of een veel kleinere rol spelen.

a. Embedded Figures Testen

Shah en Frith spraken in 1983 in een artikel over een 'islet of ability' (een enkel, uitzonderlijk vermogen) bij kinderen met autisme: veel beter dan controlegroepen konden zij vormen aanwijzen die verborgen ('embedded') waren in een grotere betekenisvolle figuur. Om deze predispositie nader te onderzoeken herhaalden wij dit experiment van Frith met de Children's Embedded Figures Test (CEFT) maar nu bij kinderen met een milde vorm van ASD (PDD) waarbij we een andere klinische groep, namelijk kinderen met het Tourette Syndroom (TS), en kinderen zonder ontwikkelingsproblemen (TD) als controlegroepen namen (zie Hoofdstuk 2). De testresultaten bevestigden onze hypothese niet: alle drie de groepen waren even snel en even accuraat in het ontdekken van de verborgen vormen. Hieruit concludeerden wij dat de taak te eenvoudig was en dus de complexiteit en sensitiviteit miste om verschillen in prestatie te kunnen laten zien.

In een tweede experiment (Hoofdstuk 4) maakten we aanvullend gebruik van een moeilijker versie van deze taak: de Embedded Figures Test (EFT) voor volwassenen, en includeerden we ook kinderen met ernstiger cognitieve en gedragsproblemen (autisme). Opnieuw kwamen er met de CEFT geen groepsverschillen naar voren, maar dit was wel het geval met de EFT: de gecombineerde ASD-groep presteerde beter dan de controlegroep (56% tegenover 48%

correcte scores) bij gelijke tijdscores. Binnen de ASD-groep en na correctie voor leeftijd en IQ, presteerden de kinderen met de meest ernstige vorm van ASD (autisme) beter (60% correcte scores) dan hun leeftijdgenootjes met mildere symptomen (55% correct) en de controlegroep (47% correct).

Met andere woorden, kinderen met ASD zijn iets beter in het ontdekken van vormen die deel uitmaken van een groter geheel, mits de complexiteit en sensitiviteit van de taak voldoende is om plafondeffecten te vermijden. Zelfs de jongens met een milde vorm van ASD bleken minder door de context te worden afgeleid; ze presteerden op een niveau dat lag tussen dat van de kinderen zonder ASD en dat van de kinderen met ernstige ASD. Dit komt overeen met de bestaande opvatting dat kinderen met autisme globale structuren niet of minder goed waarnemen en details beter.

b. Visual-completion-taak

Met de EFT worden vooral vaardigheden van een hoger cognitief niveau getest, terwijl visual-completion-taken meer basale perceptieve processen toetsen. Reeds met vier maanden stelt ons visuele systeem ons in staat objecten als compleet waar te nemen, zelfs als deze feitelijk alleen maar gedeeltelijk zichtbaar zijn. In onze waarneming worden objecten die deels bedekt zijn door het visuele systeem veranderd in de gewaarwording van meer volledige, complete objecten. Dit cognitieve proces wordt 'visual completion' genoemd. Wij onderzochten (Hoofdstuk 5) of de tendens bij kinderen met een milde vorm van ASD (PDD) om meer detailgeoriënteerd waar te nemen op een lager cognitief niveau zou leiden tot een verminderde integratie (visual completion) vergeleken met zich normaal ontwikkelende kinderen. Op het moment van deze studie was er eerder nog geen onderzoek gedaan naar visual completion bij kinderen, laat staan bij kinderen met ASD.

In navolging van eerder visual-completion-onderzoek bij volwassenen, gebruikten we een 'primed-matching' paradigma, waarmee op een indirecte manier wordt onderzocht hoe verschillende vormen worden waargenomen. Hierbij wordt gebruikt gemaakt van het 'priming effect' (PE): door eerst kortdurend een vorm te laten zien (prime),wordt het aansluitend onderscheiden van deze zelfde vorm versneld, ten opzichte van de tijd die het kost om dezelfde vorm te onderscheiden zonder prime. Een dergelijk PE treedt ook op als na priming twee identieke vormen worden getoond die gelijk zijn aan de prime. Door de aangeboden (target) vorm te manipuleren, namelijk door verschillende vormen aan te bieden en deze gedeeltelijk te bedekken, hoopten wij verschillen in basale visuele waarnemingsprocessen aan te tonen.

In Hoofdstuk 5 werden twee experimenten beschreven. De resultaten van het eerste experiment (zie Figuur 5.4) lieten effecten van priming zien na bedekte (local) en onbedekte (global) primes bij volledige testparen (cirkels). Hoewel dit erop wijst dat in beide groepen de 'visual completion' even goed verliep, gold dit alleen voor de eenvoudige vormen (cirkels). Zodra er meer complexe vormen werden aangeboden (mozaïekvorm) vonden we een veel geringer PE in de PDD groep in vergelijking met de controlegroep.

Bij de complexere vormen (zie Figuur 5.5) die we in het tweede experiment aanboden, leverden de globale en bedekte primes in beide groepen een vergelijkbaar patroon op. Ook nu lieten beide groepen 'global completion' zien, met andere woorden, alle kinderen gebruikten de contextuele informatie van de aangeboden vormen, maar de resultaten onthulden ook drie verschillen tussen de groepen. Ten eerste was het PE bij de controlegroep voor de globale en ook de bedekte primes op de globale testparen ongeveer even groot, terwijl het effect van de bedekte primes op de globale testparen bij de PDD groep minder sterk was. Op de tweede plaats liet de controlegroep een PE zien bij local primes op local testparen, terwijl dit bij de groep met PDD vrijwel niet het geval was. Ten slotte, in de controlegroep 'remden' local primes sterk de PE's bij globale testparen, terwijl dit in de PDD-groep nauwelijks het geval was. Daarentegen 'remden' globale primes nauwelijks de PE op lokale testparen, terwijl dit bij de PDD-groep sterk het geval was.

Op basis van de resultaten van beide visual-completion-experimenten kwamen wij tot de conclusie dat, in vergelijking met kinderen zonder ontwikkelingsstoornis, zelfs op het basale visuele waarnemingsniveau jongens met PDD er meer moeite mee hebben ongewone/niet-verwante of complexe vormen waar te nemen.

II VISUOMOTORIEK

De vraag bleef of deze atypische stijl van visuele informatieverwerking effect heeft op de manier waarop kinderen met PDD hun motorische handelingen plannen en coördineren. Om deze vraag te beantwoorden gebruikten we drie taken: (1) de Complexe Figuurtest van Rey (Rey CFT), (2) een volgtaak (tracking) en (3) de Developmental Test of Visual-Motor Integration (VMI) en vergeleken we de uitkomsten van 12 jongens met PDD opnieuw met de resultaten van evenzoveel kinderen met het Tourette Syndroom (TS) en zich normaal ontwikkelende kinderen (TD).

a. De Complexe Figuurtest van Rey

In Hoofdstuk 2 onderzochten we of het kopiëren en uit het hoofd reproduceren van een complexe figuur (Rey's Complexe Figuur; zie Figuur 2.2) een meer detailgeoriënteerde cognitieve stijl bij kinderen met milde ASD-symptomen (PDD) zou laten zien. In vergelijking met de twee controlegroepen behaalden de jongens met PDD in de kopieerconditie maar vooral in de uitgestelde (recall-) condities, waarbij ze de figuur uit het hoofd moesten reproduceren, lagere scores voor organisatievermogen en hogere scores voor detailgerichtheid. Hoewel in de recall-condities alle drie de groepen ongeveer 75% van de detailelementen wisten te reproduceren, presteerden de TD- en de TS-groep beter qua reproductie van structurele elementen waar de jongens met PDD geen onderscheid bleken te maken tussen de structurele en detailelementen; zij lijken de structurele elementen als aparte onderdelen te zien zonder enige samenhang.

De resultaten van deze studie toonden duidelijk aan dat, anders dan hun leeftijdsgenoten zonder stoornis, jongens met PDD bij het tekenen van een complexe figuur een detailgerichte strategie hanteren en minder gebruik maken van context en structuren (of deze misschien minder of niet zien).

b. Tracking-taak

Hermelin en O'Connor (1970) lieten in 1970 kinderen met autisme een 'tracking' taak uitvoeren waarbij ze een uitgefreesd spoor dat ze één keer wel en één keer niet konden zien met een pen moesten volgen. Omdat dit onderzoek nog nooit gerepliceerd was en wij meer zicht wilden verkrijgen op de effecten van een afwijkende visuele waarneming op de motoriek (zie Hoofdstuk 3), herhaalden wij hun onderzoek waarbij we de originele taak enigszins aanpasten. Gebruikmakend van vier verschillende tracés van toenemende complexiteit, lieten wij jongens met PDD, TS en de controlegroep met een elektronische pen (zonder inkt) sporen die in dunne plexiglasplaten waren uitgefreesd traceren, waarbij ze de tracés al dan niet konden zien.

In overeenstemming met wat Hermelin en O'Connor (1970) in hun groep autistische kinderen opmerkten, zagen wij dat de jongens met PDD in de 'zichtconditie' de tracés niet sneller wisten af te leggen dan de gecombineerde controlegroep, maar dat dit wel en in hoge mate het geval was in de 'geblindeerde' conditie. Hoogst opmerkelijk was de bevinding dat de volgorde van de condities er zeer toe deed. De controles presteerden veel beter in de geblindeerde conditie als ze het spoor eerder in de zichtconditie gevolgd hadden, terwijl de

PDD-groep daar helemaal niet van profiteerde, althans bij de minder moeilijke tracés en in mindere mate bij het moeilijkste spoor. Ook waren de jongens met PPD sneller dan de controles op de gemaskeerde tracés.

Gezien de opmerkelijke wijze waarop zij de tracés wisten te volgen, lijkt het erop dat kinderen met PDD voldoende hadden aan de haptische informatie en de visuele informatie in hoge mate negeerden. Blijkbaar maken kinderen met milde ASD veel minder gebruik van visuele input bij het plannen of bijsturen van hun bewegingen.

c. The Beery-Buktenica Developmental Test of Visual-Motor Integration (VMI)

We hebben onze groepen ook de VMI afgenomen, waarbij de kinderen gevraagd werd steeds complexere geometrische figuren te kopiëren (motorintegratie), te vergelijken (visuele perceptie), en te traceren (motorcoördinatie; Hoofdstuk 3). De VMI-scores van de jongens met PDD waren significant lager, hetgeen aangeeft dat hun visuomotorisch integratievermogen duidelijk slechter was dan dat van de twee controlegroepen. Onverwacht vonden we een dergelijk verschil niet voor de onderdelen die de visuele perceptie en de motorcoördinatie toetsen. Als de cognitieve stijl van kinderen met PDD inderdaad leidt tot een grotere focus op details dan op het gehele (te reproduceren) plaatje, dan zal het vormen van een representatie van deze figuur in het geheugen bemoeilijkt worden, hetgeen wellicht hun lagere scores verklaart.

Daarnaast vonden we het opmerkelijk dat de jongens met PDD steevast sneller tekenden dan de controlekinderen (gemeten over alleen de correcte trials) ongeacht de moeilijkheidsgraad van de aangeboden figuren (dus ook al bij het kopiëren van enkel lijnen).

Hoewel ons onderzoek er niet op was gericht de achtergrond te onderzoeken van de hoge tekensnelheden die de jongens met PDD wisten te bereiken, kunnen we op grond van wat we bij de VMI en eerder bij de trackingtaken zagen afleiden dat bij kinderen met PDD de visuomotorische controle in die zin beperkt is, dat zij minder steunen op visuele informatie om hun bewegingen te controleren en bij te stellen, wat hen tijd kan besparen waardoor hun bewegingssnelheid hoger wordt.

DISCUSSIE

In Hoofdstuk 6 vergeleken we de resultaten van de vier studies en opperden op basis daarvan dat de deficiënte manier van visuele informatieverwerking een van de belangrijkste factoren is die de afwijkende visuomotorische prestaties van kinderen met ASD verklaren en dat deze 'atypische' visuele perceptie niet alleen een grote rol speelt op een hoger cognitief niveau, maar, gezien hun matig vermogen tot visual completion, ook hun functioneren op een meer basaal niveau kenmerkt.

De afwijkende visuomotorische prestaties die we hebben waargenomen in onze groepen met milde en ernstiger ASD duiden op een atypische of deficiënte visuele informatieverwerking die wordt gekenmerkt door een detailgeoriënteerde cognitieve stijl die eerder door Frith (1989) en Happé en Frith (1996; 2006) was beschreven en aangeduid als zwakke (centrale) coherentie. Met hun uitvoerige overzichtsartikel en een eigen studie zaaiden White en Soldaña (2011) echter twijfel over de evidentie van de centrale coherentietheorie die mede was gebaseerd op de (C)EFT (verborgen figurentaken). Met onze studie (Hoofdstuk 4) toonden wij aan dat deze taken wel degelijk benut kunnen worden om zwakke coherentie aan te tonen. Deze taken kunnen subtiele maar kenmerkende verschillen in prestatie zichtbaar maken, mits men er voor zorgt dat de moeilijkheidsgraad ervan voldoende hoog is voor de controlegroepen.

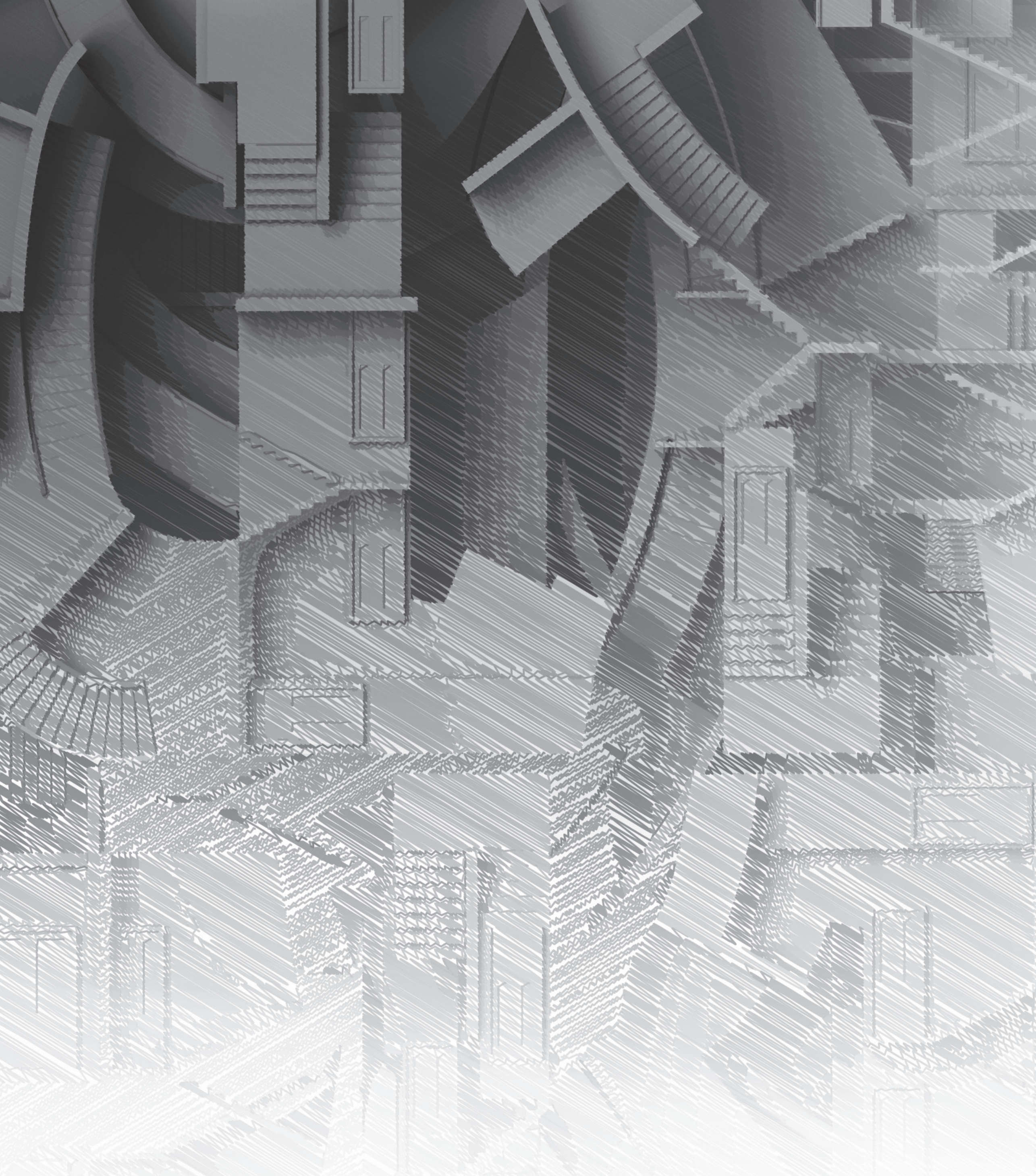
Recent formuleerden Lawson, Rees en Friston (2014) hun 'aberrant precision account of autism', een unificerende theorie over ASD die ze baseerden op het vermogen van het brein om accurate voorspellingen te genereren of te coderen ('predictive coding'). Zij stellen als hypothese dat, gezien de rol die dit coderen speelt in de dynamische regulatie van onze waarneming, ons handelen en ons sociaal gedrag, afwijkingen in de nauwkeurigheid van dergelijke coderingen ten grondslag liggen aan de sociale en niet sociale bijzonderheden die zo typerend zijn voor ASD. Dezelfde beperkingen zouden mogelijk ook andere neuropsychiatrische syndromen verklaren. In het laatste hoofdstuk van dit proefschrift bespraken we onze 'visual completion' resultaten in het licht van deze theorie.

Conclusie

Aangezien de groepsverschillen die wij aantroffen vooral kwantitatief van aard waren, kunnen we concluderen dat de processen die ten grondslag liggen aan de afwijkingen of tekorten die kinderen met milde en ernstiger symptomen van ASD vertonen, in de aard hetzelfde zijn. Onze bevindingen rechtvaardigen dan ook de praktijk om alle kinderen met stoornissen binnen het autismespectrum een soortgelijke behandeling en begeleiding aan te bieden, hetgeen met de introductie van de DSM-5 inmiddels ook algemeen wordt erkend.

De afwijkende visuele perceptie die we hebben vastgesteld bij onze kinderen met ASD beïnvloedt hun visuomotoriek op velerlei en typerende wijze. Hun waarneming verschilt niet alleen op een hoger, maar ook op een lager, basaal cognitief niveau van dat van kinderen die een normale ontwikkeling doormaken.

Het besef dat, net als hun leeftijdsgenoten met ernstige vormen van ASD, ook kinderen met minder ernstige symptomen ertoe neigen om op een fragmentarische en detailgeoriënteerde manier te werk gaan, zal niet alleen de kinderen zelf helpen beter om te gaan met de gevolgen van hun stoornis, het zal ook hun ouders, mantelzorgers, begeleiders en behandelaars in staat stellen de zwakke kanten van deze kinderen beter te begrijpen en hun sterke kanten te benutten. Kennis van welke aspecten van hun waarneming zijn vertraagd, afwijken of deficiënt zijn, en tot welke afwijkingen in hun ontwikkeling, compenserende strategieën en aanpassingsmechanismen dit leidt, zal ons in staat stellen alle kinderen met ASD beter te ondersteunen in hun dagelijks leven. Het voorkomt overvraging en biedt aanknopingspunten om hen te helpen optimaal om te leren gaan met de wereld om hen heen waardoor hun kansen en kwaliteit van leven zullen toenemen.



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Dankwoord

Bij het afronden van mijn proefschrift, kijk ik licht verwonderd terug naar de ruime tijd die dit ondernemen heeft geduurd, en de wisselende omstandigheden waarin ik in die periode als kinder- en jeugdpsychiater werkzaam ben geweest. Dit werk doe je niet alleen, zonder vele anderen kan het nooit tot stand worden gebracht. Iedereen die wel betrokken is geweest, maar die ik hier niet expliciet noem, wil ik graag laten weten dat ik hen heel erkentelijk ben.

Mijn dank gaat in de eerste plaats uit naar de kinderen en ook de ouders van alle proefpersonen voor hun persoonlijke bijdrage: ook al leken de taken soms wel computerspelletjes, ze moesten altijd volgens de regels, en helemaal tot het einde worden uitgespeeld. Uli Stibane en Koos Lukkien, beiden lid van de Raad van Bestuur van Karakter (kinder- en jeugdpsychiatrie) wil ik hartelijk danken. Jullie wilden de kinderpsychiatrie in Oost-Nederland op de kaart zetten, en steunden mijn initiatief van harte. Ik herinner me nog concreet hoe ik vanuit Zetten op een floppydisk en in hard copy mijn eerste artikel indiende. Paul Spronken, Gerton Heyne en Marie-Louise van der Kruis, bestuursleden van Reinier van Arkel en ook Henrie Henselmans en Rita van de Wouw, directieleden van Herlaarhof, ook jullie steun was altijd ruim en enthousiast: ik won de eerste Reinier Award die jullie hadden ingesteld, en kreeg vleugels. Toen wilde ik toch de verworven onderzoeksdata in artikelen omzetten, ook al had ik er eigenlijk weinig tijd voor.

Leden van de manuscriptcommissie, Bert Steenbergen, Hilde Geurts en Jan Buitelaar. Jullie reageerden meteen en positief op de vraag dit proefschrift te beoordelen, dank daarvoor.

Mijn promotor Harold Bekkering, hoofd van het Donders Instituut. Ik heb me al die afgelopen jaren gesteund gevoeld met jou in de achtergrond. Jij herkent heel goed hoe vruchtbaar de link is tussen basaal onderzoek en de klinische situatie, en bouwt bruggen tussen beide geledingen. Dit onderzoek is daar een voorbeeld van en wil ook bijdragen aan die band.

Wouter Hulstijn, mijn eerste promotor, wat kan tekenen en schrijven allemaal laten zien? In jouw handen is het een onuitputtelijke bron van onthullingen over geheugenproblemen, effecten van geneesmiddelen, schizofrenie, eetstoorissen, en ook ontwikkelingsstoornissen als autisme. Beste Wouter, jij pakte mijn vraag op, en maakte er een onderzoeksproject van. Na het eerste artikel verbond jij je vast aan deze onderneming, en brachten we uren samen door, op het laatst “achter de slangen”. Jij bracht me de liefde bij voor het scherp verwoorden, en de betekenisvolheid van een simpele taak. Op het snijvlak van fundamentele research en klinische psychiatrie onderzochten we wat de werkelijkheid is achter de uiterlijke verschijningsvorm van gedrag. Jarenlang vormden wij samen met, toen nog studenten, Pieter van den Broek, Chantalle Lankveld, Christine Trimbos, Nicole van den Bercken, en later Ivonne Wauben, Anja Vinck, Sanne Laurijsen en Inge Vermeulen een bedrijvig team dat de dataverzameling realiseerde, discussies voerde, en maaltijden genoot. Zo werden zij ook tot afstuderen begeleid. Later vormden wij dit elan om tot de werkgroep Neurocognition and Developmental Disorders. Al haar leden wil ik danken voor de deskundige scherpte in het debat, hun bevlogenheid en het plezier in onderzoek dat ze inbrachten. Ad Smitsman, Ralf Cox, Jan-Pieter Teunisse en, de helaas jong overleden, Erik van Loosbroek wil ik met name noemen. Mijn speciale dank gaat uit naar Tessa de Wit en Rob van Lier. Jullie brachten me in contact met modern visuoperceptieve research en lieten zien hoeveel clinici kunnen winnen in een samengaan met fundamenteel onderzoek.

Hooggeleerde collega, Jan Rotteveel, jij stond helemaal aan het begin van wat nu dit proefschrift is, toen we samen met Angela van der Pijll en Ria Nijhuis de polikliniek voor gedragsneurologie startten. Zo'n derdelijns poli kon niet zonder onderzoek meende jij. Ria wist dit al heel snel om te zetten in een promotie en later hoogleraarschap. Angela en jij bogen zich met mij over de vraag wat Centrale Coherentie nou precies was, en hoe je daar visuomotorisch onderzoek naar zou kunnen doen. Angela suggereerde taken en testen, en jij bevroeg me bij jou thuis kritisch wat de onderzoeksopzet was, en hoe verder te gaan. Het maakte niet uit of je wel of niet thuis was: ik werd altijd even hartelijk ontvangen. Zonder jou nam je vrouw Els de honneurs heel vanzelfsprekend en voortreffelijk waar. Dank daarvoor! Angela van der Pijll, jouw enthousiamse te zoeken wat er nu precies moeizaam verloopt in het functioneren van de kinderen die de poli voor gedragsneurologie bezoeken, werkt heel aanstekelijk. Je betrokkenheid is nooit verminderd, en nog steeds ben jij het hart van deze poli. Je treedt niet graag naar voren, maar nam toch op je mijn paranimf te zijn.

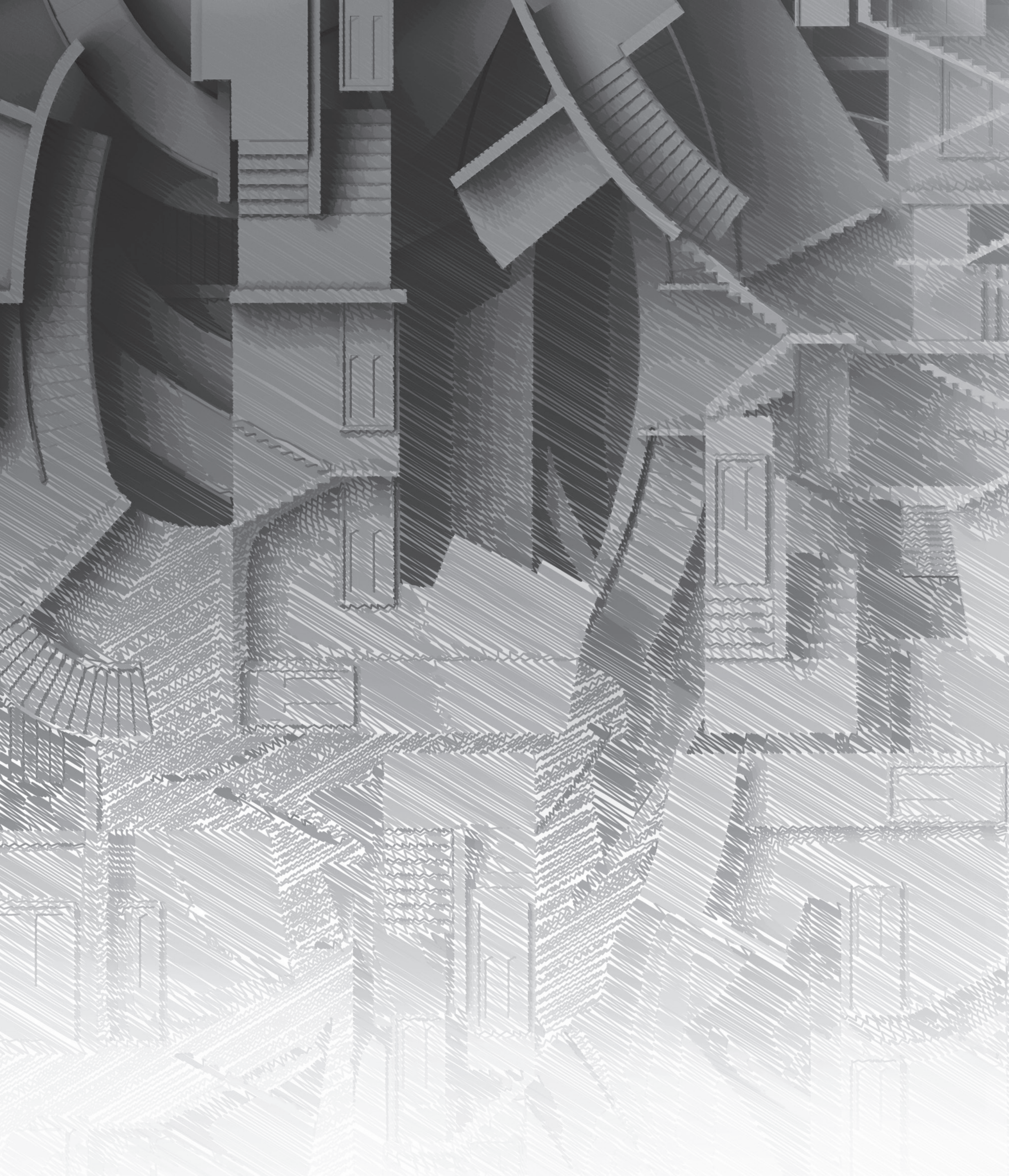
Hanneke Meulenbroek-van der Meulen, een andere kracht in de achtergrond. Jij bouwde mijn woorden niet alleen om in echt Engels, jij onderzoekt ook de connotaties van begrippen in een vakgebied dat niet het jouwe is, maar je wel goed kent. Zo kruiste je graag de degens met mij, met heldere teksten als resultaat.

Lieke, mijn enige dochter, de andere paranimf. Jij kan goed organiseren, en bent een master in planning and supply management. Maar bovenal waardeer ik je hartelijkheid, en bewonder ik je moed: niet alleen durfde jij in Zuid-Afrika van de Blaukrans Brug te bungeejumpen, je nam het ook op je om je vader in deze situatie van promoveren tot steun te willen zijn.

Pieter, Gerard en Willem, veel hebben jullie ervoor over om met zijn allen hier samen te zijn: geen tentamen of werkverplichting houdt jullie tegen. Zo lijkt dit avontuur ook weer een beetje op onze boys trip naar New York: niemand van ons weet wat we tegenkomen, maar het wordt zeker iets waar we samen lang over zullen spreken.

Gerry mijn lief. Je bracht me nooit ontbijt op bed, was vaak kritisch over de voortgang van dit project. Maar heel goed weet ik hoe zeer je me altijd steunde op jouw manier: ik word er nog steeds door getroffen hoeveel goeds me met jou is overkomen.

Lieve mam, nu ben ik ook nog aan het promoveren. 'Onze pap' zou hebben geglunderd – zeker weten.



Curriculum Vitae

Wim Schlooz werd geboren op 16 maart 1951 te Venlo. Na de middelbare school (St. Thomascollege, Venlo, 1969) studeerde hij medicijnen in Nijmegen (1979). De psychiatrie leerde hij kennen in Huize Padua (Boekel, NB) waar hij ook de opleiding begon. Aansluitend specialiseerde hij verder in het Academisch Ziekenhuis Groningen (1984, prof. dr. W.K. van Dijk en prof. dr. J.M. Minderhoud) tot psychiater en volgde aldaar ook zijn scholing in de kinderneurologie (drs. R. le Coultre en dr. C. Begeer) en kinderpsychiatrie (prof. drs. C. Rümke). In 1985 begon hij zijn werk als kinder- en jeugdpsychiater in Herlaarhof (Vught, NB), onderdeel van de Reinier van Arkelgroep. Al snel was hij daar docent bij de opleiding tot psychiater voor het onderdeel Kinderpsychiatrie.

Hij stond aan de wieg van de Therapeutische Gezinsverpleging Zuid, via welke voorziening ernstig verwaarloosde en/of mishandelde kinderen in pleeggezinnen werden behandeld, en was consulent in het Instituut voor Doven, en MKD de Kleine Cauw.

Tijdens zijn werkzaamheden bij Het Instituut voor Psychiatrische Zorg (1993–1995) zette hij een polikliniek op in Oss. Aansluitend werkte hij in het Academisch Centrum voor Kinder- en Jeugdpsychiatrie Oost Nederland (prof. dr. P. de Chateau; ACKJON, nu Karakter), verzorgde de kinderpsychiatrische consulten binnen het UMC St. Radboud, alsook in Werkenrode, de Open Cirkel en het toenmalige Pedologische Instituut. Met prof. dr. Jan Rotteveel, startte hij de polikliniek Gedragsneurologie voor kinderen. De eerste stappen in onderzoek werden hier gezet. Met dr. Aad Verrips wordt deze polikliniek nog steeds voortgezet.

In 1999 werd hij afdelingshoofd van de adolescentenkliniek in Zetten, en tevens plaatsvervangend opleider voor de kinderpsychiatrie (opleider prof. dr. R.J. van der Gaag). Consulten voor het Jeugd Justitiële Instituut de Otto Gerard Heldring Stichting en ook de Stichting Neerbosch koppelde hij aan zijn taak. Hij schreef mee aan de Multidisciplinaire richtlijn (kindergeneeskunde) 'Apparent life threatening event' (ALTE, 2006) vanuit de kinderpsychiatrie.

Hij initieerde de werkgroep Neurocognition and Developmental Disorders (2002–2010), alwaar studenten en onderzoekers op dit terrein elkaars onderzoek bespraken. Ook delen van wat later zijn proefschrift werd over de visuomotoriek bij kinderen met autismespectrumstoornissen, werden hier bediscussieerd. Zowel Karakter alsook zijn latere werkgever Reinier van Arkel (Herlaarhof) boden hem de gelegenheid om naast zijn klinische werk ook te publiceren.

Vanaf 2005 werkt hij opnieuw in Herlaarhof (Vught, NB), nu als 1e Geneeskundige; vanaf 2010 is hij alhier ook opleider voor de kinder- en jeugdpsychiatrie. Zijn aandachtsgebieden zijn neurocognitie en gedrag, en ook traumagerelateerde problematiek.

Wim is getrouwd met Gerry van Zon. Samen hebben ze vier kinderen: Pieter, Gerard, Lieke en Willem.

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ISBN 978-94-6284-034-8

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